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NEOPLASMS OF VASCULAR ORIGIN IN THE MEDIASTINUM

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PRIMARY neoplasms of vascular origin in the mediastinum are rare. This article reviews the cases so far reported in the literature, and reports 3 new cases which present some interesting features.

INCIDENCE

Seybold, McDonald, Clagett, and Harrington (1949) made the first collective review of these tumours, in which they mainly discussed their pathology. They could find only 14 cases, to which they added 3 more cases from the Mayo Clinic.

The real incidence of these neoplasms in relation to other mediastinal masses is very difficult to estimate. However, the widening scope of thoracic surgery, together with the increasing use of mass radiological surveys, has led to the discovery and treatment of many such cases. Altogether, 66 cases have been mentioned in the literature. Of these, tumours originating in the pericardium (9 cases) and the cases belonging to the hæmangiopericytoma group (3 cases) are discussed separately for convenience. Of the remaining 54 cases, only 34 are included for review in *Table I*, together with the 3 cases here described, these being the cases which have been reported in sufficient detail. Twenty-one other cases, which were briefly described or referred to by different writers while reviewing their series of mediastinal tumours, are enumerated in *Table II*. The cases described by Watson and Diamond (1947) and Schorr, Braun, and Isaac (1954) were excluded from *Table I* because of doubt about their final diagnoses in the authors' reports.

Age Incidence.—No age seems to be exempt; the youngest patient was a premature infant (Doderlein, 1938) and the oldest was a 61-year-old patient (Hosoi and Stewart, 1931). Only 6 cases were over the age of 40 years, but it is of significance to note that 6 cases were below one year of age.

Sex Incidence.—Both sexes seem to be equally affected, 20 cases being in males and 17 in females.

Incidence of Malignancy.—Ten cases were found to be malignant and 25 were benign. In 2 cases the tumour was reported as arousing suspicions of malignancy. The highest incidence of malignancy was in the third and fourth decades where 7 out of 14 cases were malignant.

Location in the Mediastinum.—The group of vascular tumours is stated to occur in all parts of the mediastinum with equal frequency (Seybold and others, 1949). However, in the cases reviewed in the present study, it was found that the anterior mediastinum was much more frequently affected (26 cases), more noticeably so in benign cases. The tumour not uncommonly reaches such considerable dimensions as to make it difficult to describe it as located in one part or the other. Most masses bulge radiologically to one side of the chest, though occasionally into both pleural cavities. Only in 7 cases was the tumour mainly posteriorly located. Two of the reported tumours originated in the diaphragm muscle.

DIAGNOSIS

It is difficult to establish the correct diagnosis of hæmangioma before thoracotomy is performed. There was only one case in this series (Heuer, 1924) in which the correct pre-operative diagnosis was reached, after histological examination of a needle-aspiration biopsy of a malignant mediastinal tumour that had eroded the ribs posteriorly. In the case reported by Adams and Bloch (1944), hæmangioma was considered the most likely diagnosis when a biopsy of a related mass in the neck was examined. This points to the importance of searching for a cutaneous or subcutaneous swelling, particularly in the cervical region in a patient with a mediastinal shadow of a doubtful nature. Actually, 7 of the reviewed cases had a benign hæmangioma protruding from within the chest. In Stewart's case (1888), a malignant tumour eroded through the sternum producing a pulsating tumour mimicking an aneurysm. In Brindley's case (1949), there were multiple glomus tumours in the right groin and leg, which were of the same nature as the mediastinal tumour. Carlson and Adams (1952) reported a huge posterior chest wall hæmangioma which extended into the posterior mediastinum as well as into the medullary canal producing hemiplegia. Inasmuch as the mediastinal mass in this case was evidently an extension from the chest wall tumour, it was not included in the present series. Owing to the predominantly anterior position in the

Table I.—REVIEW OF CASES OF BLOOD-VESSEL TUMOURS IN THE MEDIASTINUM

AUTHOR AND YEAR	SEX AND AGE	CLINICAL PICTURE	RADIOGRAPHY	TREATMENT	RESULT	PATHOLOGY
Stewart, 1888	M. 32	Cough, palpitation 1 yr. Very painful pulsating swelling over sternum 2 mth. Œdema of face and right arm		Rest and iodides for ? aortic aneurysm though negative history of syphilis	Died after developing dysphagia and ankle œdema	Angiosarcoma in anterior mediastinum infiltrating heart, S.V.C., aorta. Metastases in lung
Shennan, 1914	F. 23	Hæmoptysis 5 yr. Multiple hæmorrhages. Enlarged spleen		Deep X-ray treatment over spleen	Died, terminal mesenteric thrombosis	Hæmangioma, benign, in anterior superior mediastinum. Marked proliferative activity. Angioma in skin, lung, spleen, ovarian cysts
Kott, 1922	F. 36	Pain in chest. Dyspnœa, progressing rapidly, 5 mth.	Shadow filling left chest, more intense in 3 mth., displacing heart to right		Died of exhaustion	Angiosarcoma with invasion of anterior chest and metastases
Heuer, 1924	M. 33	Pain in right chest 5 yr. Cough and dyspnœa 1 yr. Loud systolic murmur 'extra-cardiac' over right chest posteriorly	Large mass in right upper chest mainly anterior. Not pulsating	Radium treatment, no response. Thoracotomy, inoperable. Decompression at 6th, 7th, 8th ribs posterior. Tumour pulsating	Died 4 mth. later	Malignant pleural endothelioma
Heuer, 1924	F. 17	Swelling below left scapula, 8 mth., became larger and painful. Pulsating and systolic bruit over it. Aspirate biopsy showed endothelioma or sarcoma	Mass left lower posterior mediastinum. Destruction of 9th and 10th ribs	Excision of mass and pleura and ribs over it	Well after 2 yr.	Endothelioma (pleural). No metastases malignant but pleura over tumour showed plaques and masses of tumour
Lilienthal, 1927	F. 4	Lump medial to right scapula when 8 mth. old. Recent paresis of both legs progressing to flaccid paralysis	Mass in right lower chest extending to left above the heart. Partial erosion of adjacent ribs and vertebræ. Mass present when 8 mth. old	Evisceration of tumour without opening the pleura. Treated with Cooley's fluid	Well 10 yr. after. X-ray chest: Normal paraplegia persisted	Angiosarcoma, highly malignant invasive properties
Brustalon and Parere, 1927	F. 51	Cough, pain in chest, increasing. Firm pulsating mass in right supraclavicular region. Hæmoptysis. Hoarseness of voice	Mass in right superior mediastinum anterior, pushing trachea to the left		Died before treatment	Endothelioma, malignant. Metastases on diaphragm markedly proliferative
Winkelbauer, 1929	M. 29	Pain in right chest. Slight dyspnœa and cough	Mass in right chest anteriorly, pushing heart to left	Excision readily accomplished	Died 2 mth. after operation of recurrence and metastases in both lungs	Tumour excised: cavernous hæmangioma, benign. Post-mortem: Malignant hæmangio-endothelioma
Hosoi and Stewart, 1931	F. 61	Cough for 1½ yr. following attack of pneumonia, hoarseness of voice, and sense of choking. Retrosternal dullness across upper chest	Mass in right superior mediastinum behind the œsophagus. Not pulsating. X-ray negative 7 mth. before		Died in hospital before treatment	Malignant endothelioma, encapsulated. No metastases
Pettersen and Romanus, 1933	M. 5 mth.	Lump, band-shaped, over left shoulder and left scapula, bluish colour; grew rapidly in 2 mth. Cough and stridor. Terminal dyspnœa and cyanosis	Upper mediastinum as broad as heart shadow. Mass in anterior. No increase in size in 3 mth.		Died after several weeks in hospital	Cavernous hæmangioma at site of thymus, growing ouside chest wall on left and into neck
Doderlein, 1938	M., prem. 7 mth.	Autopsy findings			Died during delivery	Hæmangioma diffusely affecting diaphragm

Table I.—REVIEW OF CASES OF BLOOD-VESSEL TUMOURS IN THE MEDIASTINUM—*continued*

AUTHOR AND YEAR	SEX AND AGE	CLINICAL PICTURE	RADIOGRAPHY	TREATMENT	RESULT	PATHOLOGY
Duvoir, Picot, Pollet, and Gaultier, 1939	F. 16	Asthmatic symptoms. Hæmoptysis once, not repeated. Congenital macrodactyly, multiple lipomata. Patient syphilitic	Shadow in left hilar region 8 yr. before, gradually increasing in size; filled left lower chest posteriorly	Thoracotomy; lung showed angioma of left lower lobe besides mediastinal angioma	Tumour unchanged after 4 yr. Died of progressive asphyxia	Lipo-angioma in mediastinum. Angiomata of left lung, liver, spleen. Associated lipomata in stomach
Adams and Bloch, 1944	M. 34	Cough persisting after a respiratory infection. Irregular movable mass in neck	Mass in left upper chest anterior. Multiple rounded opacities, more dense peripherally	Excision incomplete	Well 4 yr. later	Cavernous hæmangioma with phleboliths and bone-marrow elements
Bergstrom, 1945	F. 26 d.	Laboured breathing and cyanosis, getting worse 2 wk. Recent cough and vomiting	Right diaphragm obscured by dense triangular shadow at cardiophrenic angle. Mass is anterior. No movement of right diaphragm		Died 2 wk. after onset of symptoms	Benign hæmangioma. Capillary and cavernous replacing muscle of diaphragm
Bergstrom, 1945	F. 36 d.	Black and blue areas of skin on hips and back of hands. Regurgitation of bloodstained mucus			Died about 1 wk. after symptoms	Cavernous hæmangioma involving capsule and intralobar connective tissue of thymus and tissues around
Seybold, McDonald, Clagett, and Harrington, 1949	M. 27	Pain in the chest for 3 wk. Loss of weight of 8 mth. duration	Mass anterior to left lung hilum, pushing trachea and heart to right. Not pulsating. Increased in size during last 4 mth.	Exploration revealed mass filling of mediastinum and invading pericardium. Biopsy only	Developed superior vena caval obstruction, ? cerebral metastases. Died after 2 mth.	Hæmangio-endothelioma grade I malignancy with invasive properties, ? metastases
Seybold and others, 1949	M. 18	Mass radiography showed a shadow in right chest. Pain in lumbar region since age of 10, radiating in last 2 yr. into abdomen. ? Spondylolithesis. Lost 10 lb. in weight	Mass in right chest in relation to D.9–10 vertebræ. Erosion of D.10 vertebra. No change in size in 6 mth.	Excision. Tumour found to invade sympathetic ganglion. Post-operative radiography	Well 3 yr. after treatment	Hæmangio-endothelioma with invasive nature
Seybold and others, 1949	M. 59	Mass radiography finding. No symptoms	Mass well circumscribed posterior to hilum of right lung	Excision	Well 8 yr. after operation (Ellis and others, 1955)	Benign hæmangioma
Thomas and Chesser, 1950	F. 18	Mass radiography showed a shadow in the chest. Vague distress in chest for 1 yr.	Mass in region of left hilum anteriorly	Excision		Cavernous hæmangioma with aggregations of lymphoid cells in stroma
Horst and Beatty, 1951	M. 20	Mass radiography pick-up. Dullness over left base	Mass occupying most of left chest anteriorly. Not pulsating, mediastinum central	Excision difficult in region of the pedicle		Hæmangiofibroma
Hirschfield, 1951	F. 3 mth.	Soft swelling in right supraclavicular region. Respiratory distress, more marked on compressing mass	Shadow across apical part of right chest, displacing mediastinum to left continuous with neck mass	Aspiration of neck cysts, later thoracotomy and aspiration of mediastinal cysts. Post-operative irradiation	Slight improvement. Died 2½ mth. after operation	Hæmangioma of neck and anterior mediastinum with spindle cell matrix
Schlumberger, 1951	M. 26	Incidental autopsy finding				Fibrolipohæmangioma in anterior mediastinum cone
Perasalo, 1952	F. 48	Cough and dyspnœa after ? influenza. Dullness over left lower chest	Shadow in left lower and mid-zones, anterior, well defined, ? pulsating. X-ray 2 yr. before negative	Excision of mass with capsule. Adhesions to lung	Well 1 yr. later	Hæmangioma with suggestion of malignancy
Maggi, Barousse, and Cardeza, 1952	F. 24	Dyspnœa and cough, worse in 2 mth. Dysphagia, cyanosis. Telangiectases on face and anterior chest wall	Bilateral shadow across the mediastinum, lobulated, anterior, more bulging on right side	Treated with nitrogen mustard for a possible lymphoma	Died	Hæmangio-endothelioma, capillary and cavernous

Table I.—REVIEW OF CASES OF BLOOD-VESSEL TUMOURS IN THE MEDIASTINUM—*continued*

AUTHOR AND YEAR	SEX AND AGE	CLINICAL PICTURE	RADIOGRAPHY	TREATMENT	RESULT	PATHOLOGY
Grimes, Raphael, and Stephens, 1953	F. 31	Shadow in mediastinum discovered on routine X-ray 4 yr. before; only slight increase in size recently	Mass well-circumscribed in superior mediastinum posteriorly and on the right	Excision of well-encapsulated but adherent tumour		Cavernous hæmangioma
Sebesteny, 1953	F. 47	Pain in left scapular region increasing in last 5 mth. Shortness of breath	Mass in front and below left hilar region	Excision with capsule with no difficulty		Cavernous hæmangioma
Maurer, 1953	F. 38	Shortness of breath. Pain in the chest	Mass above and anterior to left hilum, shown by angiocardiography to be extravascular	Thoracotomy in prone position inadequate. Sternum-splitting thoracotomy: excision	Well 5 wk. later	Endothelioma in region of great vessels. No signs of malignancy
Keegan, 1953	F. 18	Loss of weight and anorexia. Epigastric pain	Mass anterior to left hilar region; smaller mass below it. Angiography showed pressure on superior vena cava	Thoracotomy: biopsy only. Post-operative irradiation done elsewhere	Condition unchanged 7 mth. later	Benign hæmangioma invading heart and possible invasion of a lymph-node
Emery and Doxiadis, 1953	M. 8	Cough, dyspnœa on slight exertion, and loss of appetite for 3 wk. Hard lump in right side of neck. Prominent veins over right chest. Horner's syndrome	Opacity filling almost all left chest. Mediastinum pushed to right. 4 yr. before there was opacity of left upper zone diagnosed as pneumonia	Following bronchoscopy cough and stridor became so severe as to necessitate tracheotomy	Died 2 hr. after tracheotomy	Benign hæmangioma with compression of the big veins of neck and trachea, which were mainly posterior to tumour
Jenny and Ulsperger, 1954	M. 26	General fatigue. Pain in chest for 5 wk.	Homogenous shadow in right hilar region, smaller rounded shadow in left hilar region. Both anteriorly placed	Excision of right lung and right mass. Excision of left mass, sternal split approach	Died 3 mth. after operation	Malignant hæmangio-endothelioma invading right lung and lymph-node metastases
Valle, 1954	M. 23	Mass radiography finding	Mass in lower anterior mediastinum, bulging to the left side	Excision	Well 18 mth. after	Hæmangioma (benign) adherent to diaphragm and other mediastinal tissues
de Maria, 1954	M. 1 yr. 6 mth.	Bluish compressible mass in left supraclavicular region. Sudden increase in size 1 mth. before on violent coughing	Shadow obscuring upper ⅔ of left lung field, displacing mediastinum to right side	Excision without difficulty. Left upper lobe atelectatic		Hæmolymphangioma extending into the neck
de Maria, 1954	M. 42	Asthenia for 2 mth. Acute attack of cough, expectoration, and pain in the chest	Mass in anterior and middle mediastinum bulging on left side between left upper lobe, bronchus and diaphragm	Excised totally. Found adherent to pericardium and great vessels		Hæmangio-endothelioma, benign in nature
Ellis, Kirklin, and Woolner, 1955	M. 9	Shadow in mediastinum found on routine X-ray. Easy fatigability	Mass in upper zone of left lung field anteriorly confluent with mediastinum, possibly pulsating	Total excision, dissected with difficulty	Well 2 yr. after operation	Cavernous hæmangioma, benign in nature
Balbaa and Chesterman, 1957. Present series: *Case* 1	M. 11	Pain in left chest, transient. Hæmoptysis for few days	Shadow in superior mediastinum bulging into both sides. Obstruction of superior vena cava	1952: thoracotomy and biopsy. 1955: excision incomplete	Well 10 mth. later	Benign hæmangioma. Marked infiltrating edges
Case 2	M. 7 wk.	Difficulty in breathing. Mass in right neck and axilla	Shadow in superior mediastinum, pushing trachea to left	Attempted excision after test irradiation 1000 r.	Died during operation	Hæmolymphangioma
Case 3	M. 21	Pain in left chest, 5 mth. Rapid weight-loss of 25 lb.	Mass bulging posterior to left hilum	Incomplete excision after test dose of 400 r.	Died 17 mth. after operation	Angiosarcoma with infiltration and recurrence

mediastinum of this group of tumours, especially the benign ones, the possibility of a tumour being an angioma ought to be considered whenever a patient presents with an anterior mediastinal shadow.

In 7 cases the tumour was merely an incidental radiological finding in essentially asymptomatic persons. This observation confirms the value of mass radiological surveys in the diagnosis of intrathoracic lesions.

The commonest presenting features were pain in the chest, cough, and dyspnœa, which are equally common in all mediastinal tumours and cysts and are non-specific. Almost all asymptomatic patients had a benign tumour. As previously mentioned, a mass in the axilla, root of the neck, or over the thoracic cage was noted in some cases. Among the rarer symptoms were Horner's syndrome due to pressure on the upper dorsal sympathetic nerve (Emery and Doxiadis, 1953), hæmoptysis due to an associated vascular tumour in the lung or arteriovenous fistula (Shennan, 1914; Duvoir, Picot, Pollet, and Gaultier, 1939), or occurring in the absence of any lung lesion, as in *Case* 1. Hæmatemesis due to associated vascular tumours in the intestines was found in Shennan's and Duvoir and others cases.

Table II.—Cases of Mediastinal Tumour not described in Detail

Author	Year	No. of Cases	Diagnosis
Watson and Diamond	1947	1	Angiofibroma; Schwannoma?
Harrington	1949	10	Endothelioma (5 cases) Hæmangio-endothelioma (5 cases)
Brewer and Dolley	1949	1	Endothelioma
Tutien	1949	1	Angiofibroma
Svanberg	1952	1	Hæmangioma
Gross	1953	1	Hæmangioma
Talman	1953	1	Hæmangioma
Peabody, Strugg, and Rives	1954	1	Hæmangio-endothelioma
Schorr, Braun, and Isaac	1954	1	Hæmangioma? Lymphangioma? Vascular teratoma?
Ellis, Kirklin, and Woolner	1955	2	Angiosarcoma (2 cases)

RADIOLOGICAL FINDINGS

Blood vascular tumours usually appear as well-circumscribed shadows even in some malignant cases. Although more commonly located in the anterior mediastinum, they have been reported in almost all other subdivisions of the mediastinum. In one of Bergstrom's 2 cases (1945), the tumour which diffusely infiltrated and replaced the right hemidiaphragm showed as a triangular shadow in the right cardiophrenic angle. Although this appeared to be fixed to both the diaphragm and the right heart border, it did not move on respiration (Van Alstyne, 1945).

Erosion of related ribs was observed in 2 cases (Heuer, 1924; Lilienthal, 1927), and erosion of the vertebral bodies in 2 cases (Lilienthal, 1927; Seybold and others, 1949). These were all malignant cases.

Fluoroscopy of the patient's chest showed transmitted pulsations in only a few cases. In Perasalo's case (1952) these gave the impression of fluid in the mass. Expansile pulsations were suspected in Ellis, Kirklin, and Woolner's case (1955). On the other hand, masses which were found to be actively pulsating during thoracotomy could not be recognized during fluoroscopic examination.

Angiocardiography proved rather disappointing as it failed to reveal the vascular nature of these tumours in those cases in which it was done. Probably this is due to the fact that the circulation of blood may be rather sluggish in the cavernous tissue of the neoplasms, which may also be partially obliterated by thrombosis. It could, however, reveal extravascular pressure on the cardiac border (Dotter and Steinberg, 1949) or the great vessels as in *Case* 1 (*Fig.* 679), in which it showed almost complete occlusion of the superior vena cava. It must be pointed out that the number of cases in which angiocardiography has been used is still too small to permit any conclusions to be reached.

Tomography and kymography, where used, added very little to the information obtained by standard radiographs and fluoroscopy. Phleboliths in a mediastinal shadow were demonstrated only in a doubtful case of hæmangioma. Possibly they occur more often, but require a special fine technique to be visualized. Their presence, however, indicates merely that the tumour is vascular and not necessarily unresectable; therefore the finding of phleboliths should not obviate thoracotomy as suggested by Schorr and others (1954).

PATHOLOGY

Neoplasms of blood-vessel origin have always been a source of controversy in the literature, concerning their origin, classification, and nomenclature. The basic structure of this group of tumours is composed of tubes, lined by endothelia, which are enclosed within sheaths of connective-tissue fibres of varying density. A simple classification, based on embryological and histological differences, such as that adopted by Stout (1945) has much to recommend it.

The majority of the benign members of this group of tumours is now considered developmental in origin. Whether they arise from sequestrated embryonic unipotent angioblastic tissue, as orginally postulated by Ribbert (Bergstrom, 1945) or from excessive development of more or less normal vessels, as suggested by Ewing (1940) is still disputed. They have been referred to by some pathologists as vascular 'hæmatomas', while the remaining tumours in this group are considered to originate as a true neoplastic process affecting the vessel walls and the supporting connective tissue. The differentiation between them is rendered difficult by the possible occurrence in the first type of disorderly richly cellular areas, which may be poorly differentiated, particularly in infants. Besides, the vessels may mingle with the surrounding tissues, giving the deceptive appearance of infiltration (Willis, 1952).

Stout (1945) classifies this 'true neoplastic' group into two main types, each of which can be benign or malignant. First, there are the rare but interesting hæmangiopericytoma tumours which arise from the pericytes of Zimmerman, and these will be discussed separately. Second, the hæmangioendothelioma tumours in which the endothelial cells

play the main role; they are arranged usually in more than one layer around vascular lumina. Excessive proliferation of atypical endothelial cells, together with very freely anastomosing channels characterize the malignant cases. Probably most of the malignant tumours of blood-vessels could be included in this group. Hæmangiopericytoma and hæmangio-endothelioma tumours are sometimes difficult to differentiate and only with the aid of silver reticulin stains and perhaps tissue cultures has this become possible.

Multiple Angiomatous Tumours.—Seybold and others (1949) mentioned as a criterion for inclusion in their series of primary vascular tumours in the mediastinum the absence of co-existent similar tumours in the lung, to exclude the possibility of the mediastinal tumours being metastatic in lymph-nodes. This, however, did not prevent them from including Shennan's case in their series. Willis (1952) states that "... while malignant angioblastic neoplasms certainly do occur and sometimes yield blood-borne metastases, the great majority of cases of multiple angiomatous formation are multifocal lesions, frequently developmental in origin". This can explain the multiplicity of benign tumours in the mediastinum as well as in other sites, e.g., liver, spleen, lung, etc., as described in Shennan's case. This has long been referred to in the literature as a "benign metastatising" hæmangioma, an entity which is denied by Stout (1945) and Ackerman (1953). The fact that the patient was young and had had evidence of the presence of vascular tumours for several years, besides their presence mainly in reticulo-endothelial territories, argues strongly in favour of a multicentric origin. On this basis, the case described by Duvoir and others (1939) can be included in this group of tumours.

Tumour Characteristics.—At thoracotomy, an angioma commonly presents as a well-defined encapsulated soft neoplasm which has a dark plum colour and may be pulsating. But quite often it forms an extensive mass that almost fills an entire hemithorax, while the edges mingle with adjacent mediastinal tissues, giving the deceptive appearance of an infiltrative growth. In some cases it actually strangles some of the main vessels of the mediastinum, e.g., the aorta (Kott, 1922; Bergstrom, 1945) or the superior vena cava (Seybold and others, 1949; *Case* 1 reported here). True invasion of the lumen of the pulmonary artery was described by Shennan (1914). Transgressing the pleural barrier and infiltration of the lung was not noted except in malignant cases. Similarly, erosion of adjacent bony structures is characteristic of malignant tumours.

In some cases the nature of the tumour may not be disclosed until actual dissection of the mass is started or even until histological examination is carried out.

In the case described by Hirschfield (1951), the thin-walled transparent cysts give the impression of a lymphangioma rather than a hæmangioma, but Ewing describes large cystic dilatations filled with clear fluid as consistent with the diagnosis of hæmangioma as long as the microscopic picture is characteristic. On the other hand, some tumours actually show evidence of a mixture of lymphangiomatous as well as hæmangiomatous tissue, hence the term hæmolymphangioma which readily describes the cases of de Maria (1954) and *Case* 2 here reported.

Malignant angiomatous tumours may well be localized and encapsulated and easily shelled out at operation, although usually they tend to invade adjacent organs, erode related bones, and produce lymphatic and even blood-borne metastases. These mostly start as malignant neoplasms, but the question of malignant transformation of benign angiomata seems to be a matter of controversy. Some writers mention its occurrence as a definite hazard; Perasalo (1952) describes in his case what appears to be a recent malignant change in a tumour in which the infiltrative growth is early. Pack and Miller (1950) in their review on hæmangioma are of the opinion that malignant change is such a rare occurrence that it does not influence their treatment. On the whole, there is paucity of histological evidence to support either view in the literature reviewed.

Hæmangioma of the Pericardium.—This forms a small subgroup, of which only 9 cases were reported. Of these, only the case reported by Bencini (1936) was included in the series studied by Seybold and others (1949). These tumours were discussed in a comprehensive review by Greenberg and Angrist (1948). *Table III* includes those cases reported so far in the literature. Mention is made here only of a few of the features which may puzzle the clinician.

The patient presenting a picture of acute cardiac tamponade without a history of trauma may have an intrapericardial rupture of a pericardial angioma

Table III.—Cases of Hæmangioma of the Pericardium

Author	Year	Pathological Diagnosis
Redtenbacher	1889	Angiosarcoma
Lefas	1898	Cavernous hæmangioma
Timme	1915	Cavernous hæmangioma
Bencini	1936	Angioma
Scheideggar	1937	Malignant hæmangioblastoma
Greenberg and Angrist	1948	Cavernous angioma Malignant angio-endothelioma
Stout	1949	Benign hæmangiopericytoma
De Carlo and Lindquist	1950	Hæmangiosarcoma

(Lefas, 1898). This tendency to spontaneous rupture may be accounted for by the constant traumatization inflicted by the beating heart.

Similarly, a previously healthy individual showing unexplained rapid decompensation and venous congestion with an enlarged cardiac shadow should suggest the remote possibility of a gradually progressive bleeding from a pericardial tumour (Redtenbacher's case, 1889). In malignant cases, the increasing bulk of the tumour itself may insidiously produce a comparable picture. Small benign angiomas, however, may remain silent, and be discovered only at routine autopsy. (Bencini's case, 1936.)

Hæmangiopericytoma and Glomus Tumour.—Of this very interesting group of tumours, only 2 cases have been described in the mediastinum, by Brindley (1949), and Ferguson, Clagett, and McDonald (1954). A third case of hæmangiopericytoma in the pericardium was described by Stout (1949). Pathologically the tumour may be

soft and vascular or hard and sclerotic. It affects any age and may even be found in the newborn. It has no characteristic site in the mediastinum and may attain any size. The single characteristic feature of these tumours is the histological picture which essentially shows profuse proliferation of capillaries and endothelial buds, each of which is surrounded by a connective-tissue sheath and one or more layers of concentrically arranged pericytes of Zimmerman. Non-myelinated nerves and nerve-endings may or may not be present. Whether glomus tumours and hæmangiopericytomas are variants of the same neoplasm or are different entities is still disputed. Ferguson and others, however, conclude that " . . . according to Stout's own terminology glomus tumors might well be haemangiopericytomas with the appearance of glomus-like bodies ".

The clinical picture of Brindley's case is interesting, as the patient suffered from chest pain that had the characteristic paroxysmal nature of peripherally situated glomus tumours. Both cases were excised successfully with disappearance of the patients' symptoms. The literature does not contain any case report of a malignant hæmangiopericytoma of the mediastinum.

TREATMENT

Surgical excision is certainly the treatment of choice in benign cases owing to their possible complications. It must be realized, however, that this may be a tedious procedure because of the infiltrative nature of some of these neoplasms, and substantial blood-loss may necessitate transfusion of several pints of whole blood. The desirability of a generous exposure cannot be over-emphasized; in 2 of the reported cases a second operation was necessary to gain a better access to the tumour. This can be avoided, particularly in cases in which the mass appears to bulge into both thoracic cavities, by utilizing an anterior bilateral approach obtained either by splitting the sternum vertically, or by dividing it transversely in line with a bilateral intercostal incision.

In some cases it may be advantageous to stage the operation to obtain complete excision. In *Case* 1, part of the mass had to be left behind. In a future operation excision of this remaining portion, together with the occluded segment of the superior vena cava, replacing it with a homograft, will probably have to be carried out.

Irradiation therapy is not of much value in benign cases, as the tumour is not radio-sensitive. Besides, the patient being a child in a good percentage of cases, may suffer unnecessary damage inflicted on his growing thoracic cage by radiotherapy.

For the malignant cases, total excision is the only chance for cure, probably more so if combined with radiotherapy. The latter alone may offer the patient with an inoperable growth some palliation, though the results are far from encouraging.

Of the reviewed cases, 20 underwent operative removal of the tumour; 11 of these had complete removal, and 9 had only incomplete excision which was followed in one case by deep X-ray therapy. In 3 cases, only a biopsy was made because the tumour was considered inoperable (Seybold and others[3] 1949), or because the disease was widespread in the lungs and mediastinum (Duvoir and others, 1939). Heuer (1924) attempted decompression of the chest by excision of the overlying ribs without much success, the patient surviving only four months after operation.

Though malignant tumours usually have a poor prognosis, yet in some cases a combination of excision and radiotherapy (Seybold and others, 1949) or even surgical excision alone (Perasalo, 1952) can lead to two or three years' survival. Radiotherapy was considered rather hazardous to attempt in the latter case owing to the presence of active tuberculosis in the lungs.

It might be of interest to note that the longest survival in the malignant cases was in the case reported by Lilienthal, which was treated by evisceration of the tumour, followed by systemic injection of Cooley's fluid; the patient was reported alive and well with radiological disappearance of the tumour ten years later.

CASE REPORTS

Case 1.—A. L., a male child 11 years old, was admitted to the City General Hospital, Sheffield, on account of a large shadow in the superior mediastinum. He had no complaint on admission, but 10 weeks previously he complained of pain in the chest which was sharp and more noticeable on inspiration. This gradually improved without treatment. Seven weeks later he developed a fever of moderate degree accompanied by mild hæmoptysis which lasted only 5 days. There was history of whooping-cough complicated by pneumonia, scarlet fever, chicken pox, and bronchopneumonia, all before the age of 4.

On Examination.—A pale thin child with slight cyanosis. No lymph-nodes were palpable and there was no clubbing of fingers. The tonsils were enlarged. Pulse and temperature were normal. Chest examination revealed a pigeon-breast deformity and Harrison's sulci. There was fullness over the left infraclavicular fossa with some prominent veins and diminished percussion note over the same area. Few rhonchi were heard over the left base. The heart was normal and the blood-pressure was 110/70. No skin lesions were noted. Physical examination was otherwise normal.

Radiography.—Examination of the chest (*Figs.* 676, 677) showed an opacity confluent with the superior mediastinum and protruding to the right and to the left. It had a well-defined convex margin, and appeared to 'sit' on the cardiac shadow. In the lateral film it appeared to lie in the anterior and middle portions of the superior mediastinum. Fluoroscopy confirmed these findings. The trachea was slightly deviated to the right and slightly narrowed in the coronal plane. There was only transmitted pulsation. The shadow did not move with swallowing and barium swallow showed a normal œsophagus. The mass was assumed to be of thymic origin. Laboratory findings were within normal limits.

At Operation.—(Aug. 7, 1952, Mr. J. T. Chesterman.) Left posterolateral thoracotomy through the bed of the resected fifth rib was made. A large nodular mass of varying consistency occupied the upper mediastinum. Dissection was started by opening the overlying pleura, and freeing of the tumour was attempted but had to be given up after one hour, owing to its adherent and infiltrative nature and its extension into the right chest. No enlarged lymph-glands or metastases were encountered. A biopsy was taken and the chest was closed without drainage. The post-operative course was uneventful, the patient being discharged on Aug. 26, 1952.

HISTOLOGICAL EXAMINATION (*Fig.* 678).—A nodule composed of a network of vascular channels, varying in size from that of a capillary to that of a sinus. Some of these channels were thin-walled, with little more than two endothelial layers separating them. There was some fibrous stroma, intervening in places with occasional stromal hæmorrhages. Scanty foci of lymphocytes and fat cells were present. Diagnosis: Cavernous hæmangioma.

Post-operative radiotherapy of the tumour was suggested but refused by the radiotherapist owing to the insensitive nature of the neoplasm and the danger of the procedure on the bony structure of the thorax.

FIG. 676.—*Case* 1. Postero-anterior radiograph.

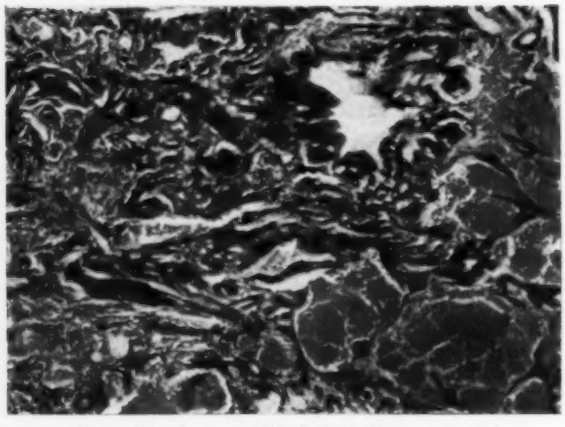

FIG. 678.—*Case* 1. Histological picture. (× 50.)

Subsequently, the boy was forced to lead a somewhat restricted life though he could swim and play football a little.

ON RE-ADMISSION.—Owing to some increase in the size of the tumour, local discomfort in the chest, and retarded growth, the boy was re-admitted on March 6, 1955.

ON EXAMINATION.—Congested jugular veins were more noticeable on the right side. Angiocardiography (*Fig.* 679) revealed the presence of almost complete obstruction at the lower end of the superior vena cava; most of the dye escaped through the azygos vein and other collateral channels. The other findings were essentially the same. It was decided to attack the tumour through an adequate exposure.

AT OPERATION.—March 28, 1955. The chest was entered this time through a transverse anterior incision, opening up the right and left second intercostal spaces and dividing the sternum at the same level with a Gigli saw. Excellent exposure was obtained and the tumour was found to extend from beyond the superior vena cava on the right to the region of the descending thoracic aorta on the left. Freeing of the tumour was very tedious, especially the posterior aspect of the central portion, due to the presence of very thin-walled venous channels around the tumour mass. There was a large communicating vessel about 1½ cm. in diameter entering probably into the right atrium, the exact anatomy being very difficult to identify. The greater portion of the tumour and its vascular connexions were excised, but ultimately a portion behind the superior vena cava and the posterior part on the left side had to be left behind when it was realized that the patient's condition would not allow further prolonged dissection. The chest was closed with underwater drainage bilaterally. Seven pints of blood were given, and the operation required about 4½ hours. The patient made a slow but uneventful recovery. Histological examination of the resected specimen showed essentially the same picture as the previous biopsy, with no evidence of malignancy. Ten months later, the patient is developing well and attempting with more success to lead a normal life.

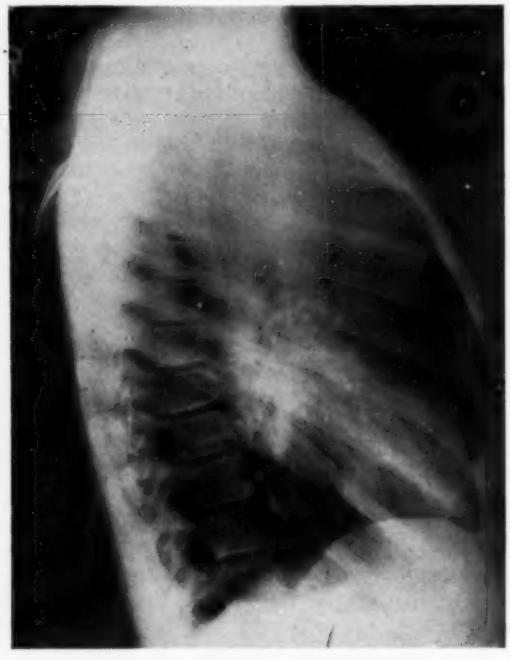

FIG. 677.—*Case* 1. Lateral radiograph.

Case 2.—L. C. B., a male infant 7 weeks old, was admitted to the University of Michigan Hospital in Ann Arbor, Michigan, on June 5, 1940. Difficulty in breathing and choking spells during feeding had occurred during the two weeks prior to admission.

ON EXAMINATION.—A well-developed baby, breathing comfortably when quiet, but showing laboured breathing while crying. The entire right side of the neck was occupied by a large cystic mass which appeared to bulge more on crying, and seemed to extend behind the clavicle into the right axilla. Rhonci were found over both lung fields.

Radiography (*Fig.* 680).—Marked enlargement of the mediastinal shadow above the heart. The trachea was sharply displaced to the left, and in the lateral view it was seen to be narrowed and displaced posteriorly. It was

felt that the mass in the mediastinum was connected to the cervical and axillary masses and it was believed that the lesion was not of thymic origin. Laboratory findings were within normal limits.

Biopsy of the axillary mass revealed a ' lymphangioma cavernosa '. The baby was therefore referred for deep

resected first rib. A large neoplasm was found protruding from the upper mediastinum and displacing the apex of the lung down to the level of the fifth intercostal space posteriorly. It was mostly solid postero-inferiorly and partly cystic anterosuperiorly. Anteriorly it seemed to enclose the superior vena cava and subclavian artery.

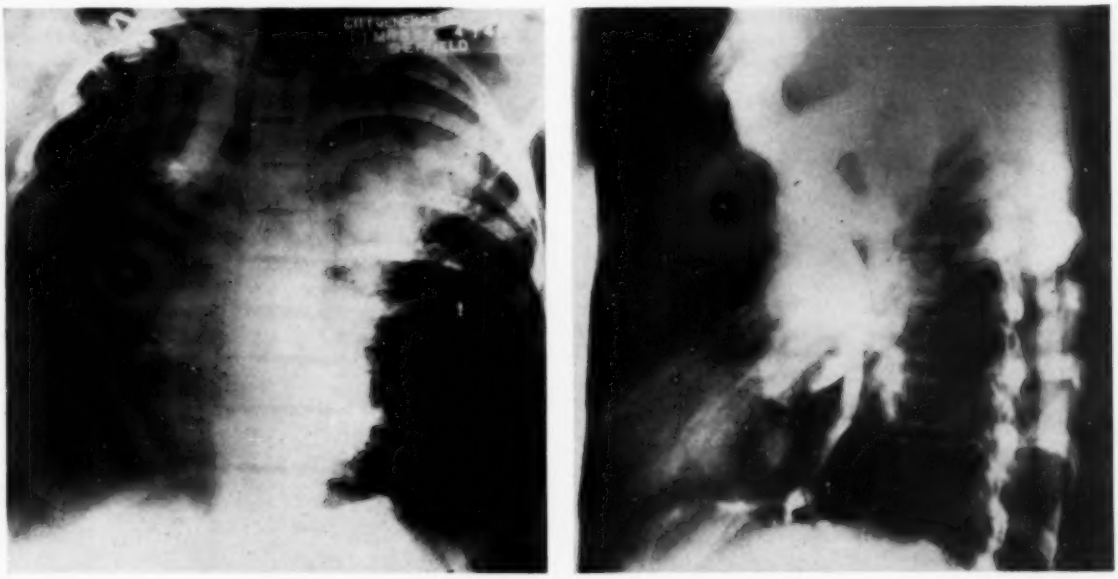

FIG. 679.—*Case* 1. Angiocardiogram showing almost complete occlusion of the superior vena cava, and large accessory vessels connecting the mediastinal veins with the tumour mass.

FIG. 680.—*Case* 2. Radiograph showing huge mediastinal tumour. Note the cervical and axillary masses.

FIG. 681.—*Case* 2. Histological picture. (× 100.)

X-ray therapy; 1000 r units at 5 exposures were given to the chest, neck, and axilla. There seemed to be no apparent response to the treatment. Because of rapid deterioration in the baby's condition, an operation was decided upon.

AT OPERATION (June 26, 1940, Dr. John Alexander).—A posterolateral thoracotomy incision was made and the right pleural cavity was entered through the bed of the

The tumour was lobulated and adherent to the various mediastinal structures and was infiltrating into the neck and axilla. During attempted excision of the tumour, the infant's condition deteriorated and he died before the operation was completed.

Autopsy revealed considerable narrowing of the trachea, and the cause of death was probably multiple segmental atelectases of both lungs.

HISTOLOGICAL EXAMINATION (*Fig.* 681).—There were areas which consisted of many large endothelial-lined spaces filled with blood-corpuscles. Smooth muscle was present in their walls. Other similar sinusoidal spaces had no blood-cells in them and small collections of lymphocytes were visible between the spaces. The tumour was interpreted as a combination of cavernous hæmangioma and lymphangioma, better named hæmolymphangioma. The tumour in the neck was predominantly a hæmangioma, whereas that in the axilla was mainly a lymphangioma.

FIG. 682.—*Case* 3. Radiograph of chest.

Case 3.—R. H., a 21-year-old man, was admitted to the University of Michigan Hospital, Ann Arbor, Michigan, on Nov. 3, 1942, with a chief complaint of pain in the posterior portion of his left chest, of 5 months' duration. This started as dull and occasional pain, but became intolerably severe and constant prior to admission. There was a 25-lb. weight loss during that period. A previous history of left pleurisy 2 years previously was obtained. This was associated with hæmoptysis, and this occurred while the patient was doing some work which necessitated exposure to silica.

ON EXAMINATION.—The pulse and temperature were normal. The blood-pressure in the right arm was 105/60 mm. Hg, and 130/90 mm. Hg in the left arm. There was absent tactile vocal fremitus anteriorly and posteriorly over the left upper chest. Breath-sounds had a bronchial quality with scattered râles over this area. There was generalized palpable lymphadenopathy and a large soft gland was palpated high up in the left axilla. Laboratory findings were within normal limits.

Radiography (*Fig.* 682).—A smooth rounded mass bulging into the left thoracic cavity at about the level of the pulmonary hilum, and pushing the trachea slightly over to the right side. In the lateral view, it proved to be posterior to the hilar region, extending toward the vertebræ and ribs, but with no apparent bone erosion. A tentative diagnosis of probable neurogenic tumour or possible lymphoblastoma was made.

A test dose of irradiation of 400 r was delivered in two anterior and two posterior parts, with no significant reduction in the size of the tumour in 6 weeks.

Bronchoscopy on Nov. 6, 1942, showed hyperæmia and œdema of the left main bronchus and compression of the bronchus below the level of the upper lobe orifice.

AT OPERATION.—Thoracotomy was performed by Dr. John Alexander on Nov. 9, 1942, through the bed of the resected fifth rib. A fist-sized tumour was found to bulge into the upper half of the left main fissure and actually invaded the lung tissue. Sharp dissection was required to free it from the lung, vagus nerve, sympathetic trunk, and from the vertebral bodies. Part of the tumour tissue, infiltrating deeply into the anterolateral wall of the thoracic aorta could not be removed, for the aorta was accidentally nicked during the dissection, but was readily sutured. The chest was closed with drainage.

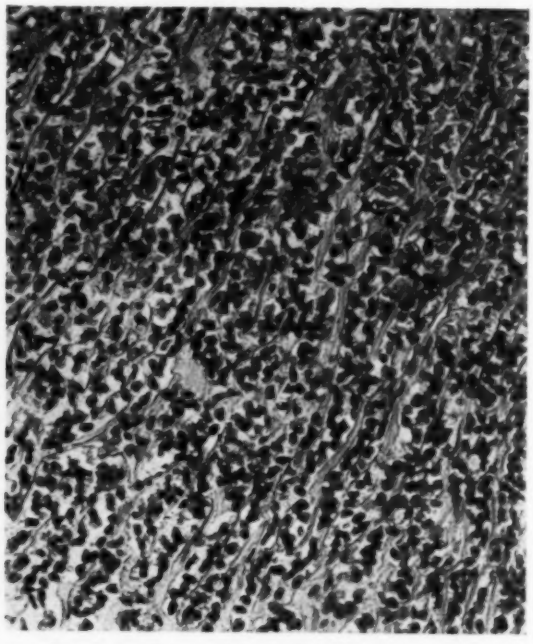

FIG. 683.—*Case* 3. Histological picture. (× 165.)

PATHOLOGICAL EXAMINATION (*Fig.* 683).—A circumscribed and partially encapsulated neoplasm consisting of polyhedral cells arranged in alveoli and columns. In some areas there were obvious vascular spaces lined by neoplastic cells. This was interpreted as an 'angiosarcoma' and the neoplastic cells as of endothelial origin.

The post-operative course was uneventful. Post-operative irradiation of the tumour area with 3000 r was given and the patient was discharged on Dec. 16, 1942. Following discharge, the patient's condition and chest radiographs were satisfactory until November, 1943, when he began to complain of weakness and weight loss and hoarseness of voice. X-rays revealed recurrence of the tumour. His condition gradually deteriorated until he died on March 5, 1944.

SUMMARY

1. The literature concerning the neoplasms of blood vascular origin located in the mediastinum is reviewed. Sixty-six cases were found, but only those reported in sufficient detail are summarized in table form to facilitate future reference.

2. Three previously unreported cases which present some interesting features are reported in detail with photomicrographs.

3. The clinical picture and the value of different diagnostic procedures are discussed in enough detail to help in making a pre-operative diagnosis.

4. Some controversial points in the pathology are discussed.

5. Operative removal is the only effective treatment to avoid the occurrence of possible complications.

Acknowledgement.—Thanks are due to Dr. Cameron Haight, head of the Department of Thoracic Surgery (University of Michigan, Ann Arbor, Michigan) for allowing the publication of the 2 cases from his department.

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POST-TRAUMATIC THROMBOSIS OF THE INTERNAL CAROTID AND VERTEBRAL ARTERIES AFTER NON-PENETRATING INJURIES OF THE NECK

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THE syndrome of spontaneous thrombosis of the internal carotid artery without any preceding trauma is now well recognized and there are numerous references to it in the literature. Gurdjian and Webster (1953) carried out arteriographic studies in a series of patients suffering from acute hemiplegia and found that in 30 cases the cause was a thrombosis of the internal carotid artery.

There have also been described a number of cases of thrombosis of the internal carotid secondary to penetrating wounds of the neck or cheek. As would be expected, the great majority of these cases were seen during the last two world wars (Makins, 1919; Caldwell and Hadden, 1948).

The syndrome of thrombosis of the internal carotid artery following a blunt non-penetrating injury to the neck is, however, only rarely seen. Schneider and Lemmen (1952) were able to find 5 cases in the literature, to which they added 2 cases of their own. Caldwell (1936) described one case and a further case was reported by Clarke, Dickson, and Smith (1955). This makes a total of 9 recorded cases, to which I wish to add a further example. I have been unable to find a reference in the literature

Table I.—RECORDED CASES OF THROMBOSIS OF THE INTERNAL CAROTID AND

	VERNEUIL 1872	GRECO 1935	CALDWELL 1936	ERIKSON 1943	NORTHCROFT AND MORGAN 1944
Age	Not stated	23	16	28	Young soldier
Nature of injury	Railway accident	Blow on left side of face and neck	Struck side of head	Blow on right side of neck and head	Caught round neck by rope
External evidence of injury when first seen	Contusion vertex skull	Abrasions left side of face	Induration and tenderness left side of neck	Not stated	Swelling left sternomastoid. Abrasions left side of neck
State of consciousness soon after injury	Confused but conscious	Dazed. Able to ride bicycle	Dazed but able to talk	Lethargic after a few hours	Alert but amnesic. Able to walk
Order of onset of neurological signs and symptoms	Right hemiplegia and coma in next 24 hr.	Headache and blurred vision in few hours. Coma and right. hemiplegia in 16 hr.	Semi-comatose in 6 hr. Right hemiplegia in 10 hr.	Left hemiplegia after few hours	Right-sided Jacksonian fits → right hemiplegia next day. Semi-coma
Latent interval between injury and onset of signs	Less than 24 hr.	Less than 16 hr.	6 hr.	A few hours	Approx. 24 hr.
Whether craniotomy performed	No	Yes. N.A.D.	No	No	Yes. N.A.D.
Other diagnostic procedures	—	Lumbar puncture. N.A.D.	—	Right carotid angiography	Ventricular puncture. N.A.D.
Radiographs of skull	—	Negative	Negative	Negative	Negative
Pulsation of vessels on affected side of neck or face	Not stated	Not performed	Carotid pulse diminished	Not stated	Not stated
Clinical diagnosis	Cerebral hæmorrhage	Middle meningeal hæmorrhage	Intracranial hæmorrhage	Thrombosis right internal carotid artery	Intracranial hæmorrhage
Specific treatment	None	None	None	None	None
Result	Died	Died	Died	Died	Died
Post-mortem findings	Thrombosis left internal carotid. Softening left cerebral hemisphere	Thrombosis left internal carotid. Softening left cerebral hemisphere	Thrombosis left internal carotid extending to middle cerebral. Cerebral softening	Thrombosis right internal carotid. Large infarct right cerebral hemisphere	Dissecting aneurysm left internal carotid with thrombosis into circle of Willis. Infarct left cerebral hemisphere

to thrombosis of the vertebral artery following a non-penetrating injury, and here report such a case.

One of the main problems of these injuries appears to be in recognizing that a patient with neurological signs and symptoms, and a history of a closed injury to the neck or head can be suffering from a thrombosis of one of the main extracranial vessels supplying the brain.

In an attempt to give a clearer idea of the syndrome a tabulated summary has been made of all the recorded cases. (*Table I.*)

CASE REPORTS

Case 1.—**Thrombosis of right internal and external carotid arteries.**

A man of 42 sustained a heavy blow from a cement hopper on the right upper chest and neck at about 2.30 p.m. on Oct. 24, 1955. He was taken to hospital and examined as an out-patient. His cervical region and upper right thorax were X-rayed and the wet films interpreted as showing no evidence of gross bony injury. The patient was fully conscious and was able to give a clear account of the accident. Clinical examination was negative except for some local tenderness and abrasions over the right supra-clavicular region. He was able to dress himself unaided. He refused the offer of an ambulance to take him home and departed, as he came, in the firm's lorry.

A few hours later he was brought back to hospital in a dazed and confused state. There was, by this time, some bruising and fullness of the right anterior triangle of the neck. He had also developed a hemiplegia affecting the whole of the left side of the face, and the left arm and leg. He had become drowsy and his speech was slow, slurred, and indistinct. There was conjugate deviation of the eyes to the right, the pupil of the left eye was slightly enlarged, and both reacted to light. There was no abnormality of the fundi. The pulsation of the carotid arteries was not clearly felt on the right side, while the facial and superficial temporal pulses were definitely not palpable. A thrombosis of the right carotid arteries on the right side was considered to be

VERTEBRAL ARTERIES FOLLOWING NON-PENETRATING INJURIES OF THE NECK

CALDWELL AND HADDEN 1948	SCHNEIDER AND LEMMEN 1952 (*Case* 1)	SCHNEIDER AND LEMMEN 1952 (*Case* 2)	CLARKE AND OTHERS 1955	MURRAY 1957 (*Case* 1)	MURRAY 1957 (*Case* 2)
20	42	24	45	42	16
Blow on right side of face and neck	Blow on right side of head by a car	Blow on right side of face and neck	Blow on left side of neck	Blow on right side of neck and chest	Drawn into machinery. Blow on back of neck
Fracture right jaw. Small cut right side of neck	Contusion right supraclavicular region. Fracture right clavicle	Abrasions right side face and neck	Abrasions left side of neck. Contusion left side of head	Abrasions right clavicular region. Fracture 1st right rib	Lacerations right side face. Abrasions face, neck, and thorax
Momentarily stunned. Recovered quickly	Conscious but soon became lethargic	Semi-conscious	Fully conscious. Could describe accident	Alert. Able to converse and dress himself	Shocked, restless, semi-conscious
Semi-coma and left facial palsy in 4 hr. Left hemiplegia in 16 hr.	Semi-coma and left hemiplegia in 12 hr.	Coma and left hemiplegia during next 36 hr.	Aphasic in 3 hr. Right facial palsy and right arm paralysed in 12 hr. Right hemiplegia next day	Drowsiness and left hemiplegia developed within 6 hr. of injury	Semi-coma → profound coma in 12 hr. Signs of brain-stem lesion
4 hr.	Less than 12 hr.	Less than 36 hr.	3 hr.	4 hr.	12 hr. approx.
Yes. N.A.D.	No	Yes. Cerebral œdema	No	No	No
Ventricular puncture. N.A.D.	Right carotid angiography	Right carotid angiography	—	—	—
Negative	Negative	Negative	Negative	Negative	Not X-rayed
Not stated	Carotid pulse felt	Carotid pulse clearly felt	Normal pulsation in superficial temporal	Diminished pulsation in neck. No pulsation in facial or superficial temporal	Strong carotid pulse
Subdural hæmatoma	Thrombosis right internal carotid artery	Extradural hæmorrhage. Later thrombosis right internal carotid artery	Thrombosis left internal carotid artery	Thrombosis right internal carotid artery	Brain-stem hæmorrhage
None	Superior cervical sympathectomy	Continuous procaine block of cervical sympathetic chain	Anticoagulant therapy started on 3rd day	Anticoagulants and vasodilators on 1st day	None
Died	Recovered. Residual left hemiplegia	Died	Died	Died	Died
Thrombosis right internal carotid. Intima torn extensively and acting as a valvular obstruction	—	Thrombosis right internal carotid into circle of Willis. Marked swelling right cerebral hemisphere	Thrombosis left internal carotid with intimal tear. Left cerebral hemisphere swollen and soft. Pontine hæmorrhages	Extensive dissecting aneurysm right common carotid with thrombosis of internal and external carotids. Massive softening right cerebral hemisphere	Thrombosis of right vertebral artery. Massive softening right lobe of cerebellum. Medullary hæmorrhages

the most likely diagnosis and anticoagulant therapy was started at once. In addition, vasodilator drugs were given orally. Ten thousand units of heparin were administered intravenously, and dindevan was given by mouth. Thereafter the heparin was continued by intravenous injection every 4 hours for 48 hours and the dindevan orally according to the 'prothrombin content'.

PROGRESS.—On the evening of the day of admission he became blind in the right eye. All the superficial tendon reflexes and the abdominal reflexes were absent. The left plantar reflex was definitely extensor. There appeared to be anæsthesia of the left side of the body.

His general condition showed little change over the next two days. He remained lethargic, able to speak in a slurred indistinct voice. It was reported that he had moved his left leg, but this was unconfirmed and did not occur again. Meanwhile, the anticoagulant therapy was continued. In the early hours of Oct. 27, 1955, the patient's condition began to deteriorate. He became deeply comatose. His temperature rose to 104°, the pulse varied between 52 and 114 beats per minute, and the blood-pressure was also variable but inclined to be high. Both lower limbs and the left arm were held rigid. The plantar reflexes were now extensor on both sides; Cheyne-Stokes respirations developed. He gradually became weaker and died at 8.30 a.m. on Oct. 27, 1955.

POST-MORTEM EXAMINATION.—

Dissection of Neck.—There was some bruising over the right shoulder and the side of the neck with superficial scarification of the skin. The neck muscles showed diffuse interstitial hæmorrhage on both sides, but much more severe on the right. There was a fracture of the first right rib. Both internal jugular veins were patent and their walls showed no abnormality. Dissection of the right common carotid artery showed a significant acquired abnormality at the level of the isthmus of the thyroid gland. This abnormality consisted of an intramural hæmorrhage of the artery which had lifted the intimal coat from the outer coats and had led to gross narrowing of the lumen over a distance of almost 4 cm. At the distal end of this stretch it appeared as if the intimal coat itself had been ruptured, and beyond this point the intramural hæmorrhage became continuous with a propagated red thrombus. This thrombus had extended distally to involve both the external and internal carotids. The left common carotid artery was normal except for a few subintimal patches of atheroma. The rest of the neck, including the cervical spine, showed no abnormality.

The Head.—The scalp, skull, and meninges showed no abnormality. Inspection of the brain after removal showed distinct swelling of the right cerebral hemisphere. Section showed this to be due to a well established extensive right cerebral softening involving most of the frontal, parietal, temporal, and occipital lobes. Only a narrow rim of healthy brain-tissue remained in each of these lobes. The softening extended deeply to involve the internal capsule, the head of the caudate nucleus, and the putamen, together with the globus pallidus. The thalamus appeared to have escaped. The remainder of the brain, the cerebellum, and the brain-stem appeared to be normal.

Inspection and dissection of the internal carotid artery as it entered the skull revealed ante-mortem propagated red thrombus which had entered the circle of Willis to involve the right anterior and middle cerebral arteries and passed along each of them for a distance of a few centimetres.

Comment.—The diagnosis in this case lay between an extra- or subdural hæmorrhage and a thrombosis of the right internal carotid artery. Attention was directed to the site of injury at the root of the neck on the right side by the presence of swelling and bruising. The absence of pulsation in the branches of the external carotid and the barely perceptible pulsation of the common carotid led to a diagnosis being made of carotid artery thrombosis and allowed anticoagulant treatment to be started early. The appearance of the lumen of the common carotid at the site of injury, as described above, suggests that a tear of the intima took place which allowed blood to strip up this coat. The space made between the intima and the media then became so filled with blood as to occlude the lumen of the vessel. It was in fact a form of localized dissecting aneurysm. The mechanism of injury to the common carotid is thought to have been a direct contusion.

Case 2.—**Thrombosis of the right vertebral artery.**

A boy of 16 was admitted to hospital on the afternoon of Oct. 27, 1955, with severe injuries to the right arm. He had been drawn into a large band-saw with which he had been tampering in the absence of the regular operator, and was found lying unconscious in a pool of blood.

On admission he was very shocked and had obviously lost a considerable quantity of blood, to judge by the saturated condition of his clothes. He was semi-conscious, pale, sweating, and restless. His pupils were both small and reacted to light. He had been given ¼ gr. morphine intravenously before admission.

His right arm had been mutilated in a grotesque fashion. The revolving saw blade had split the forearm cleanly in two along its length from the fingers to the elbow joint. The cleavage had cleanly divided the palm and the wrist joint in the coronal plane showing the carpal bones in section. The split extended up the forearm dividing it in two sections to open up the elbow joint. No further bone destruction was seen although the skin laceration extended half way above the elbow. The fingers were completely macerated.

On the right side of the face there was a series of deep punctures made by the teeth of the saw.

There were friction burns over the right side of the upper part of the chest. There was no external evidence of injury to the head.

Resuscitation was started at once. He was given 2 pints of dextran, followed by 2 pints of blood under pressure, and blood continued as a drip thereafter.

AT OPERATION.—At 5.30 p.m. his condition was considered fit to allow his right arm to be amputated. Under a light nitrous oxide and oxygen anæsthetic the right arm was amputated and the facial lacerations were sutured. His condition at the end of the operation was fairly satisfactory. The blood-pressure was 115/70 and both pupils were normal.

PROGRESS.—It was noted by the night staff that "he took a long time to come out of his anæsthetic".

Next morning he became more drowsy and by noon was in deep coma. By this time his blood-pressure was 130/75 and the pulse rate 110. The right pupil had become well dilated and neither pupil reacted to light. His temperature rose to 103°. Both plantar responses were extensor and all his limbs were in a state of hypertonus. Breathing became faint and oxygen was administered with controlled respiration. A few hours later both pupils were widely dilated and fixed. The blood-pressure had fallen to 85/48, and the pulse rate was about 160 per minute. Respirations were now very feeble whenever the artificial respiration was discontinued, and soon afterwards ceased, although the circulation still continued weakly. Death took place around 5 p.m. on Oct. 28, 1955, 24 hours from the time of operation.

POST-MORTEM EXAMINATION.—There was a severe contusion of the right upper chest and two similar contused areas in the midline and overlying the sternum. There was a 3-in. laceration of the right cheek over the right mandible which had been sutured.

There was widespread bruising of the right side of the neck. The second and third ribs on the right side were fractured near the costochrondral junction underlying the severely contused area mentioned above. The right pleural cavity contained about 10 oz. of blood. The right lung was collapsed, except for an area of the middle lobe which had been torn slightly and was emphysematous. The left lung was normal. There was no sign of foreign material in the pulmonary vessels. The heart was normal.

All organs of the abdomen and pelvis were substantially normal. None showed any infarction.

The Brain.—There was massive softening of the right lobe of the cerebellum and its peduncle. The cause of this softening was found to be a thrombosis of the right vertebral artery, the thrombus extending up almost to its junction with the left vertebral and involving the right cerebellar vessels. There was a hæmorrhagic area measuring 1 in. across in the basal part of the left lobe of the cerebellum. The cut surfaces of the lower pons and upper medulla showed some blurring of the normal pattern on the right side as compared with the left. The cerebrum showed no abnormality.

Examination of the base of the skull showed the thrombus to extend well down the right vertebral artery as far as it could be traced. There was no sign of traumatic thrombosis in any of the great vessels of the right side of the neck.

Comment.—When this case was first seen attention was focused mainly on the patient's state of shock and on the mutilated right arm. As time went on it was suspected that there was an intracranial lesion, judging by the patient's drowsiness, which remained as the state of shock wore off under treatment. There were no localizing signs on the first day, and the possibility of fat embolism was considered. This was not confirmed clinically. On the day after operation unmistakable signs of a brain-stem lesion were evident with a cerebellar pressure cone and a condition akin to decerebrate rigidity. The march of events then was too rapid for decompression to be likely to succeed although this step was considered.

The diagnosis of a vertebral artery thrombosis did not occur to anyone.

It is interesting to conjecture how this lesion was caused. Unfortunately no one saw the accident, but a reconstruction indicates that the boy's right arm was drawn into the saw by the rollers on which the planks of wood are normally placed. There are two parallel upright stanchions guarding the entrance to the saw blade and these prevented his head and body being also drawn in. Thus the neck was forced strongly to the left, hard up against the pillars. This may have caused stretching of the right vertebral artery over one of the many bone projections it encounters on its tortuous course to the brain, or even against the atlanto-occipital membrane which it penetrates. It is even possible that a subluxation of the cervical spine took place as the neck was wrenched forcibly to the side with spontaneous restitution. No gross dislocation or fracture could be seen in the cervical spine although it must be admitted that a full post-mortem dissection of the vertebral column was not performed.

DISCUSSION

Pathology.—It is well recognized now that contusion or stretching of a major artery can cause sufficient damage to the coats of the vessels to lead to its occlusion by spasm or thrombosis. (Allen, Barker, and Hines, 1946.) In none of the published cases (*see Table I*) was there any visible breach of the outer wall of the carotid artery, and damage was mainly directed to the intima and media. Verneuil (1872), Greco (1935), Caldwell and Hadden (1948), Northcroft and Morgan (1944), all describe intimal lacerations with splitting of the intima and media and detachment of the inner coat. In some cases the intima was curled up and acted as a plug. The damage to the intimal lining may be a clean tear, as in the case of Clarke and others (1955), or there may be such extensive stripping of the intima from the media by the force of blood-flow in the carotid artery as to justify the description of a dissecting aneurysm with occlusion of the lumen by extravasation into the vessel wall (*Case* 1).

Intimal atheroma, which probably plays a large part in the production of the so-called spontaneous type of thrombosis, is not thought to be an aetiological factor of any significance in these cases. The patients were mostly young men with no evidence of arteriosclerotic changes. It will be noted that intimal atheroma was found in the unaffected left common carotid artery in *Case* 1. Northcroft and Morgan (1944) dissected out the carotid arteries of a large group of healthy young soldiers who had died from other causes and found patchy intimal thickening in all cases.

It was noted in *Case* 1 that the large veins of the neck were unaffected by the trauma. Platt (1930), in a description of 2 cases of thrombosis of the axillary artery due to the pressure of a crutch, commented on the absence of involvement of the axillary vein. The pressure of the large veins in the neck is very low, being between 0 and −2 mm. Hg, whereas the mean pressure in the carotid arteries is about 90 mm. Hg. There is thus a much greater resistance in the artery, which would probably render it more vulnerable to contusion or stretching.

Widespread arterial spasm following injury to the large arteries may play a large part in the initial stages of this condition. Ecker (1945) gives an account of 4 cases, with signs almost indistinguishable from those described above, caused by spasm of the internal carotid following blunt non-penetrating injuries to the neck. In his cases the spasm passed off and the neurological signs gradually disappeared. In all the fatal cases of thrombosis of the internal carotid artery the thrombus extended into the major cerebral arteries and to the circle of Willis to a varying degree, and resulted in massive infarction and softening of the homolateral cerebral hemisphere. Whether this takes place as rapidly as the clinical signs suggest is open to doubt, and probably extensive spasm of the arterial bed precedes the extension of the thrombosis.

These cases illustrate the marked difference between surgical ligation of the carotid or vertebral arteries and their obliteration by thrombosis. French and Haines (1950), in describing a fatal case after ligation of one vertebral artery, refer to the frequency with which this vessel has been tied off by others, once even bilaterally, without major complications. Lambert Rogers' figures (1949) show that the common carotid may be ligated with comparative safety, but that ligation of the internal carotid carries with it a considerable risk. He quotes 8 deaths and 12 cases of cerebral complications out of 51 cases. This is a high mortality figure, but even so does not compare with 10 deaths out of 11 cases of carotid and vertebral artery

thrombosis. The difference lies mainly in the spread of the thrombosis occluding possible channels of collateral blood supply.

Diagnosis.—From the account of the published cases (*Table I*) it will be seen that the main obstruction to the diagnosis in these cases is that the clinical picture so closely resembles that of the more common extra- or subdural hæmorrhage. Attention is apt to be focused on the cranial condition and the train of neurological signs, as the external evidence of injury is often indefinite, or complicated by other apparently more urgent injuries. When a patient is seen with an injury to the neck and subsequent neurological signs suggestive of an intracranial lesion, thrombosis of the internal carotid artery should be suspected.

In *Case* 1 diagnosis was made easier by the fact that the patient was able to give a history of a clear-cut injury to the neck and chest, and by the absence or diminution of facial, temporal, and carotid pulses when seen a few hours later. Caldwell and Hadden (1948) believe that absence of the temporal pulse on the affected side is of assistance in diagnosing occlusion of the external and possibly the common carotid artery. Schneider and Lemmen (1952), and Clarke and others (1955), however, were able to feel clearly pulsation in the carotid and also the facial and temporal vessels, in spite of thrombosis of the internal carotid. Pulsation can be transmitted to the thrombosed segment from the unoccluded part, and therefore, although it is a help to find no pulsation in the vessels of the neck, the presence of pulsation does not exclude a thrombosis. Gurdjian and Webster (1953) found no help in the diagnosis of spontaneous internal carotid artery thrombosis from pulsation of the vessels in the neck.

A further aid to the diagnosis being made in *Case* 1 was the early onset of homolateral blindness which was thought to be due to either spasm or thrombotic occlusion of the central artery of the retina. This occurrence was not observed in any of the other cases. Walsh and Smith (1952) point out that ligation of the ophthalmic artery close to the internal carotid does not affect the eyesight because of the free collateral circulation between it and the external carotid system. Homolateral loss of vision would result, however, if both the internal and external carotids on that side were affected by thrombosis, as in *Case* 1.

A craniotomy was performed in 4 of the previously recorded cases (*Table I*) as an intracranial hæmorrhage could not safely be excluded. In 3 cases a healthy-looking brain was seen and in 1 there was evidence of cerebral œdema.

Schneider and Lemmen (1952) used arteriography as an aid to their diagnosis and state that it is the most accurate method of diagnosing thrombosis of the internal carotid artery during life (*Table I*). This is probably true, but the procedure is not easy and, as mentioned by Clarke and others (1955), it is not without its hazards. It should never be employed bilaterally because of the danger of prejudicing the collateral circulation.

The diagnosis of a thrombosis of the vertebral artery following a non-penetrating injury of the neck or head is one which cannot be reasonably expected to be made during life.

Treatment.—To have any chance of success, specific treatment must be started early and this of course depends on a prompt diagnosis. Treatment should be designed to prevent the extension of the thrombotic progress, to prevent embolism, and to abolish spasm and so maintain the maximum efficiency of the collateral circulation. As soon as a diagnosis is made anticoagulant therapy should be started. This can be a difficult decision to make in cases where the diagnosis is in doubt—and this means in almost all the cases. Clarke and others (1955) state that they refrained from giving anticoagulants at once because of the possible danger of hæmorrhage into the softened area of the brain. This did not occur in *Case* 1, although anticoagulants were given for 3 days, unfortunately with no beneficial effect. Anticoagulants have been reported as giving good results in spontaneous thrombosis of the carotid by Caldwell and Hadden (1948) and they quote others who have had the same experience.

The relief of spasm of the cerebral vessels can be attempted by use of antispasmodic drugs either orally or parenterally. Tolazoline, or priscol, has lately been used intra-arterially in cases of peripheral vascular obstruction, particularly of the lower limbs, with beneficial effects. It would appear that it has a place in treating the initial spasm here, and if injection could not be given into the affected artery then perhaps injection into the common carotid on the unaffected side would be still effective in allaying the spasm of the collateral vessels. The dosage recommended is 100 mg. of tolazoline in 20 c.c. saline.

A more certain and lasting effect can be obtained by a superior cervical sympathectomy, according to Schneider and Lemmen (1952), whose only surviving case was so treated. They use the open technique to perform arteriography and if this shows a thrombosis to be present they proceed to do a sympathectomy. Poppen (1950) inserts an indwelling polythene catheter around the carotid vessels to perfuse the sympathetic chain with a procaine drip in cases where he has to ligate the common carotid as an emergency procedure.

Ligation of the thrombosed segment of the carotid vessel has been reported by Gurdjian and Webster (1953) to give good results in 15 of their 30 cases of spontaneous thrombosis of the internal carotid. This condition is obviously slower in onset and more localized to a segment of the vessel than is the more rapid propagation of thrombosis following injury. Caldwell and Hadden (1948) suggest trying to ligate the carotid above and below the thrombus in early cases. They recommend an intracranial approach for the distal ligation, but this would appear to be an heroic measure to take in the presence of thrombosis, considering Lambert Rogers' (1949) experience of ligation of the internal carotid where no thrombosis existed at the time of ligation. The operation they suggested is technically outside the scope of any but an experienced neurosurgeon. However, desperate measures must be employed for desperate conditions. Schneider and Lemmen (1952) point out that extensive thrombosis is not found in the early stages. Presumably, therefore, as suggested earlier, the first neurological signs are caused mainly by spasm, and if ligation can mitigate

this and prevent further thrombosis and embolism, then it is worth trying. If ligation is attempted, division of the vessel with excision of the segment must also be done to prevent pulsations driving the clot onwards.

SUMMARY

The 9 previously published cases of internal carotid artery thrombosis following closed injuries have been summarized. One further case has been added, and a case of traumatic thrombosis of the vertebral artery described. In the whole series of 11 cases there were 10 deaths. Attention should be directed to the possibility of a traumatic thrombosis of the internal carotid artery when a patient with an injury to the neck develops neurological signs of an intracranial lesion.

Arteriography would appear to be the most accurate diagnostic method, but it is not without its dangers. The element of spasm is emphasized, and measures to combat it are described.

I should like to record my indebtedness to Mr. R. G. Henderson for allowing me to use *Case* 1, and for his kindly and helpful criticism. I also wish to thank Dr. R. Rankin and Dr. J. F. Boyd for the post-mortem reports.

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RETICULOSARCOMA IN HASHIMOTO'S DISEASE

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SARCOMA of the thyroid gland is a rare form of neoplasm and, owing to the great difficulty in differentiating it histologically from the more common anaplastic carcinoma, its diagnosis is as a rule the subject of considerable controversy. Difficulty has occurred both with the small round-cell type of growth and also with the spindle-cell variety. Ewing (1940) from a study of his thyroid tumours concluded "that the mesoblastic origin of most of the sarcomas reported in the literature is highly improbable and that the occurrence of true sarcoma in man still requires demonstration".

Willis (1953) believes that "most of the 'sarcomas' and 'carcinoma-sarcomas' of the thyroid have been anaplastic carcinomas", but quotes the article by Kellett and Sutherland (1949) on reticulosarcoma of the thyroid gland without adverse comment. These authors described 5 cases, all occurring in elderly women. In 3 cases tumour-free thyroid tissue was available for study and each showed lymphoid hyperplasia. They were extremely doubtful, however, whether this hyperplasia comprised part of an early struma lymphomatosa.

The following two cases show the features of a reticulosarcoma arising from a thyroid gland affected by struma lymphomatosa and infiltrating into neighbouring tissues.

CASE REPORTS

Case 1.—A married woman, aged 58 years, was admitted to hospital on June 26, 1952 having had a lump in the neck for four months, during which time it had been increasing in size. A month previous to admission she had developed a dry cough.

ON EXAMINATION.—She was obese and hypertensive (B.P. 195/100) but her appearance was healthy. There was a visible goitre, both lobes of the thyroid being appreciably and uniformly enlarged and firm in consistency. In addition there was a midline swelling which appeared to arise from the isthmus and this was of a harder consistency than the rest of the gland, suggesting the possibility of malignancy. Radiographs of the chest revealed no evidence of metastases.

AT OPERATION (D. H. C. H.).—On June 27 operation was performed, and on exposure of the midline swelling this was seen to be a tumour, grey in colour, arising from the isthmus and apparently of a malignant nature, in that it was beginning to invade the overlying muscles. On further exposure the lateral lobes were seen to be uniformly enlarged, paler than normal, firm in consistency and they were well defined, showing no evidence of involvement of the surrounding tissues or other suggestion of malignancy. These appearances were in fact typical of Hashimoto's disease. In view, however, of the convincing evidence of malignancy of the midline tumour, an almost total thyroidectomy was performed, leaving only a small wedge of thyroid tissue on the right side.

Convalescence was satisfactory and, in view of the subsequent histological report, the patient was treated with radiotherapy (2950 r over a period of 21 days). While in hospital she had two attacks of severe dyspnœa and laryngoscopy showed evidence of a paralysed left vocal cord presumably caused by surgical trauma. She was discharged on Aug. 7 and, when last seen as an out-patient on Dec. 27, 1956, there was no sign of recurrence of the neoplasm. She has, however, been depressed, has had hoarseness of voice, and has suffered occasional attacks of dyspnœa and giddiness. She showed evidence of

hypothyroidism until the maintenance dose of thyroid was raised to 4 gr. per day.

THE SPECIMEN.—

Macroscopical Description.—The specimen consisted of a thyroid gland, with the exception of a portion of the right lobe, and weighed 80 g. The lateral lobes showed symmetrical enlargement and slight lobulation. In the isthmus there was a roughly spherical mass 4 × 4 × 3 cm. On section the isthmic nodule displayed a mottled dull-yellow and white appearance and its anterior part invaded some overlying voluntary muscle. Laterally this swelling merged with thyroid tissue showing similar appearances on its cut surface, although in occasional areas some translucent material could be discerned.

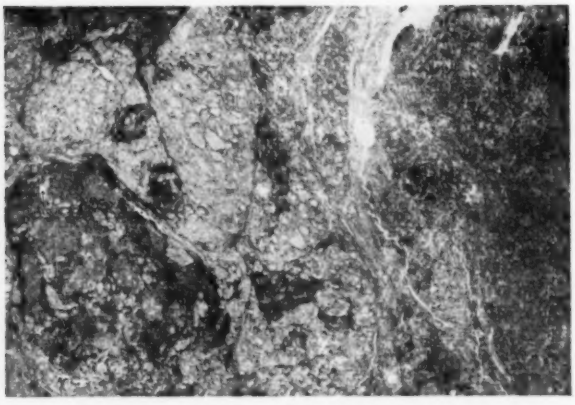

FIG. 684.—*Case* 1. Right lateral lobe, showing lymphocytic infiltration, lymphoid follicles with pale germinal centres, and sarcomatous infiltration on the right. Hæmatoxylin and eosin. (× 10.)

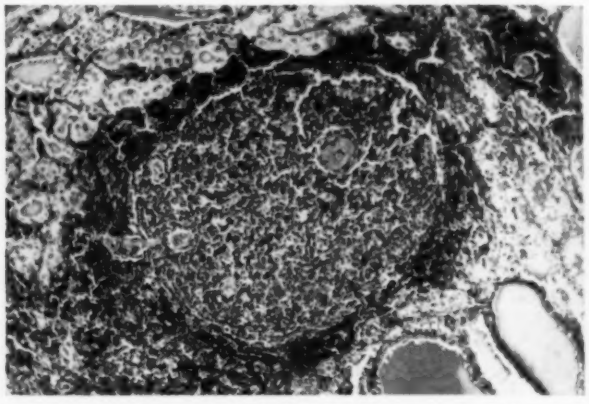

FIG. 685.—*Case* 1. Lymph follicle with large germinal centre. Hæmatoxylin and eosin. (× 75.)

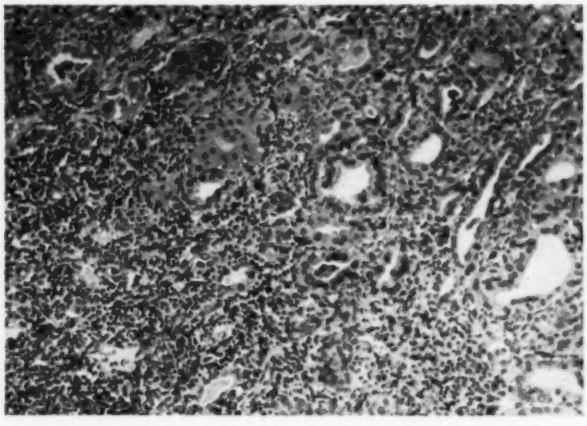

FIG. 686.—*Case* 1. Left lateral lobe. Thyroid acini composed of eosinophilic cells, infiltrated by lymphocytes and undergoing atrophy. Hæmatoxylin and eosin. (× 100.)

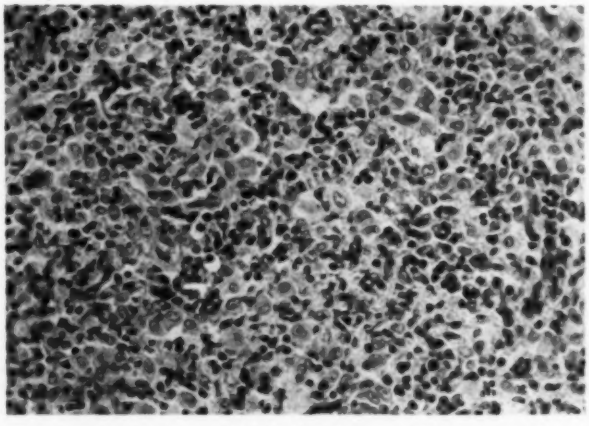

FIG. 687.—*Case* 1. Histological features of the neoplastic tissue in the isthmus. Hæmatoxylin and eosin. (× 200.)

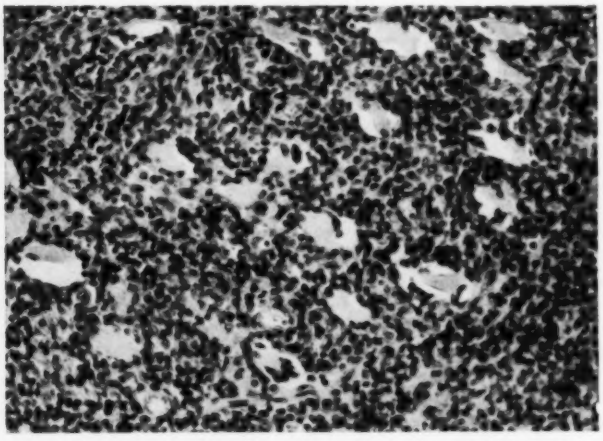

FIG. 688.—*Case* 1. Neoplastic tissue infiltrating and destroying voluntary muscle-fibres. Hæmatoxylin and eosin. (× 150.)

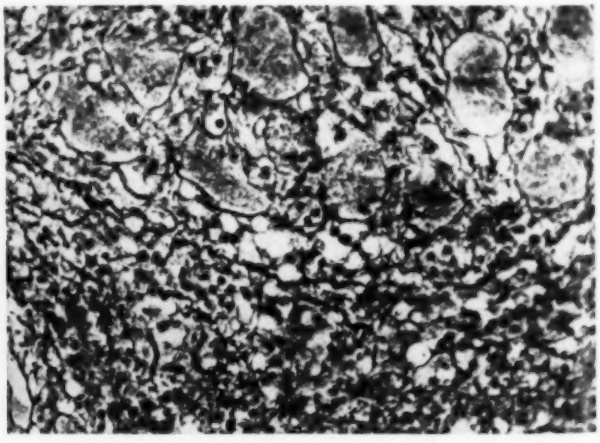

FIG. 689.—*Case* 1. Shows the reticulin network intimately associated with the neoplasm and penetrating between muscle-fibres. Reticulin stain. (× 62·5.)

Histology.—The lateral lobes are split up to some extent by coarse fibrous trabeculæ and there is a widespread cellular infiltration, composed mainly of small lymphocytes. (*Fig*. 684.) Plasma cells, histiocytes, and cells resembling reticulum cells are less numerous. In many areas the infiltrations have a follicular appearance and typical germinal centres are frequently seen. (*Fig*. 685.) The glandular tissue has been split up and destroyed to a large extent, but islands of acini remain between the cellular infiltrations (*Fig*. 686) and, even in the densest collections of lymphocytes, glandular epithelial remains can often be recognized. These remnants usually have a distinctly eosinophilic cytoplasm seen also in some other areas. The colloid content is, in most acini, either low or absent, but a small number of glands are distended by deeply eosinophilic colloid. The lining epithelium is generally of low cuboidal type.

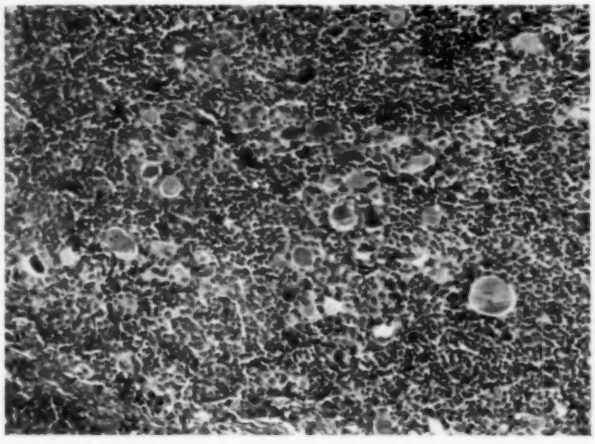

FIG. 690.—*Case* 2. First operation specimen. Heavy lymphocytic infiltration with surviving thyroid acini. Hæmatoxylin and eosin. (× 100.)

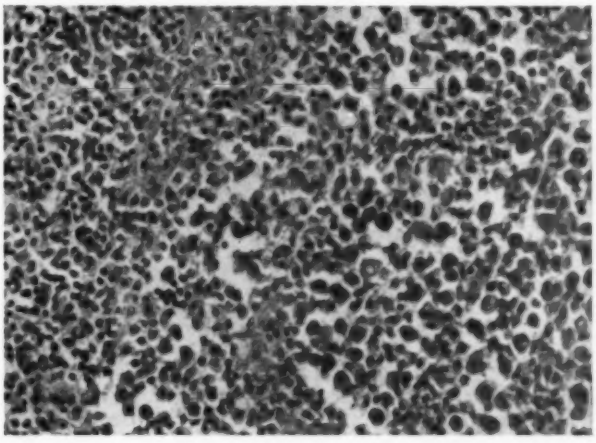

FIG. 691.—*Case* 2. Second operation specimen. Reticulum-cell sarcoma with mitotic figure. Hæmatoxylin and eosin. (× 200.)

The isthmus and the adjacent part of the right lobe show very extensive, dense infiltrations of cells which appear much more irregular than the lymphocytic infiltrations already described. They tend to be large, more variable in size, and their nuclei are more vesicular. Many resemble reticulum cells; occasional 'mirror-image' multinucleate types occur and mitoses appear more frequently. Necroses, some resulting inflammatory reaction, and phagocytosis of necrotic cells are present. The remains of acini can usually be distinguished from the infiltrating cells. The appearances in these two regions are those of a reticulosarcoma. (*Fig*. 687.) This neoplasm has infiltrated through the gland capsule and permeated and destroyed much voluntary muscle adjacent to the isthmus and the anterior part of the right lobe. (*Fig*. 688.) At the margins of the growth, columns of cells penetrating between the muscle-fibres are characteristic of neoplastic permeation and no fibrosis of muscle is present. Only a little glandular tissue remains in the isthmic mass.

Reticulin stains show a fine network intimately mixed with the neoplastic cells, and this network accompanies the infiltration into the voluntary muscle. (*Fig*. 689.)

Numerous sections have been studied, but nowhere is there evidence that this is an undifferentiated carcinoma.

Case 2.—A man aged 59 years was seen in consultation on July 21, 1952, with a history of having had a large goitre for about six years. It had increased only slowly in size until the last few months, when its rate of growth had much increased though there were no appreciable local pressure symptoms.

ON EXAMINATION.—The patient was of average build and had a healthy appearance. There was no dyspnœa and his voice appeared normal. There were no eye signs, tremors, tachycardia, undue moistness of the skin, or other signs of thyrotoxicosis. He had quite a considerable enlargement of the thyroid gland involving the left lobe and isthmus and displacing the trachea to the right. It was very firm, only slightly nodular, and not tender: it moved normally on swallowing. It was difficult to be sure whether the right lobe was enlarged at all.

FIRST OPERATION.—On July 28, 1952, the large left lobe of the thyroid was delivered without undue difficulty (J. P. H.). It had a smooth, hard surface and was not abnormally adherent to the surrounding structures. The right lobe was examined and appeared to be of average size, appearance, and consistency and it was not further interfered with. The whole of the isthmus and almost the whole of the left lobe were removed.

PROGRESS.—He had remained well for about two years when in July, 1954, he began to notice a feeling of pressure in his throat not amounting to a pain but causing some dyspnœa. This gradually increased and when he was seen in December, 1954, there was a considerable enlargement of the right lobe of the thyroid: it was very hard, rather fixed, had pushed the trachea over to the left and its lower edge could not be felt as it went down behind the clavicle. No other swellings could be felt in the neck and no metastases could be seen in a radiograph of the chest.

SECOND OPERATION (Jan. 3, 1955).—The neck was opened widely in the usual manner and the enlarged right lobe of the thyroid was exposed. On its outer side there was a well-defined capsule and separation was easy, but inferiorly behind the clavicle it was difficult. Medially the thyroid was hopelessly fixed: there was no discoverable plane of separation, the trachea and adjacent structures being infiltrated over a wide area by what was undoubtedly growth. The recurrent laryngeal nerve could not be recognized. The whole lobe was removed as far as was possible, the medial part having to be cut off the trachea and small portions of the rings being included in the specimen. The patient was left with a paralysed cord. A full course of radiotherapy (3000 r in 28 days) was given at St. Bartholomew's Hospital.

PROGRESS.—The patient has continued in moderate health but with some stridor owing to a narrow trachea and a paralysed vocal cord.

There are as yet no discoverable metastases.

THE SPECIMENS.—

First Operation.—

Macroscopical description: Mass of thyroid tissue 10·5 × 6·5 × 5·0 cm., with capsule on one surface. The other surface is raw. The tissue is pink in colour with

whitish areas up to 2·0 cm. in diameter and many dark hæmorrhagic zones.

Histology.—Surviving glandular acini are widely spaced. They are small and contain little colloid. Some acini contain cells of lymphoid type and one shows squamous metaplasia. Many of the acinar lining cells are large, with eosinophilic cytoplasm.

Between the acini there is a dense infiltration by cells of the lymphoid series, the majority of which are small lymphocytes. (*Fig.* 690.) Many lymphoid follicles, with and without germ centres, are present, but their outlines are indistinct, the component cells merging with the infiltrating lymphocytes.

In many areas proliferating reticulum cells are present.

There are some bands of coarse collagen fibre, but fine reticulin is not increased.

The appearances suggest sarcomatous change in Hashimoto's disease.

Second Operation.—

Macroscopical description: Mass from right lobe of thyroid 8·0 × 4·5 × 5·0 cm., almost completely covered by a fibrous capsule. There is a circular, rough, raw area on one aspect 3·5 cm. in diameter. Cut surface shows a fairly uniform, soft, pink tissue with an occasional fibrous trabeculum.

Histology.—Only traces of thyroid tissue now remain. There are numerous collections of lymphocytes with some germinal centres. Reticulum cells are more numerous than in the first operation specimen and vary in size. (*Fig.* 691.) Giant forms are present and a moderate number of reticulum cells are in mitosis. Also present are some foci of necrosis with associated endothelioid cells and giant cells. Among the collections of endothelioid cells are large pale cells with abundant granular eosinophilic cytoplasm, which are Askanazy cells derived from thyroid acini. These appearances are interpreted as reaction to necrosis and escape of colloid into the stroma and not secondary involvement by tuberculosis. There is marked increase in collagen fibre, which is present in coarse bundles. In places reticulin fibrils are also increased. The appearances are now those of reticulum-cell sarcoma, the picture of Hashimoto's disease having been largely obliterated.

DISCUSSION

Reticulosarcoma of the thyroid is a rare condition and occurs almost exclusively in elderly females. Graham (1933) found 11 lymphosarcomas among 222 malignant thyroid tumours; Portmann (1940) found 11 out of 200 cases; and Vaux (1937) found 1 case among 25 cases.

In earlier papers, however, the histological descriptions and illustrations have often left some doubt as to the sarcomatous nature of the tumour described. Kellett and Sutherland (1949) reviewed the literature on the subject, found 10 adequate descriptions of such tumours and added 5 more cases of their own. Dean (1949), Dinsmore, Dempsey, and Hazard (1949), Dick and Kellett (1951), and Scott (1952) have also recorded similar tumours. Lindsay and Dailey (1955) have recently described 8 cases in which they consider a lymphosarcoma had originated in the thyroid gland. In 7 of these the glands also showed the characteristic lesions of Hashimoto's disease. They believe the relationship between these two lesions to be significant. Brewer and Orr (1953), however, discussing the undifferentiated tumours of the thyroid, designated one group 'struma reticulosa' and stated that this corresponded to the condition described in the literature as reticulosarcoma. In this group, comprising 10 cases, the tumours were composed of reticulum cells and lymphocytes and some degree of fibrosis was noted. Four of the cases survived for long periods (2, 9, 10, and 16 years), and in the fatal cases 'metastases' in the gastro-intestinal tract were more frequent than could be explained by chance. They suggested that 'struma reticulosa' might be a variant of Hashimoto's struma and regarded as unproved the view that the lesions under discussion were neoplastic in the ordinary sense. However, they were unable to find any histological differences between the fatal and non-fatal cases, and they recognize the fact that malignant reticuloses of lymphoid tissue often behave differently from most tumours in regard to the sites of their metastases. In the present state of knowledge we prefer to regard the malignant reticuloses as malignant neoplasms rather than granulomatous conditions despite the fact that some of them have very long histories, that in view of their nature fibrosis may be a prominent feature, and that the metastases in fatal cases often show a predilection for certain unusual sites including the gastro-intestinal tract.

We believe that a malignant neoplasm having features identical with the lymphosarcomas and reticulosarcomas of lymphatic tissue can arise occasionally in the thyroid gland, but that this must, if possible, be differentiated from an anaplastic carcinoma and from an inflammatory lesion.

From the pathologist's point of view the main issues in *Case* 1 may be summed up as follows:—

1. Is the thyroid affected by Hashimoto's disease?
2. Is the tissue in the isthmus and part of the right lobe a neoplasm?
3. Is it a reticulosarcoma?
4. What is the significance of Hashimoto's disease in relation to the aetiology of the malignant tumour?

The appearance in Hashimoto's disease, particularly in relationship to Reidel's struma and other types of 'thyroiditis', has itself been the subject of much controversy. The appearances in the lateral lobes of the thyroid in this case are identical with those described by Hashimoto (1912), Graham and McCullagh (1931), Joll (1940), and Schilling (1945) as typical of this condition, and with those seen in 32 cases diagnosed as 'struma lymphomatosa' and studied at St. Bartholomew's Hospital. Macroscopically, the symmetrical slightly lobulated lateral lobes, some increase in the fibrous stroma, and the absence of recognizable normal thyroid tissue are characteristic. Histologically, the widespread lymphocytic infiltrations leaving only islands of thyroid acini between, and the frequent lymph follicles with prominent germinal centres, are typical of struma lymphomatosa.

It does not seem necessary to enter into the controversy as to whether Hashimoto's disease is a distinct entity, separable from the other varieties of lymphocytic infiltration and lymphoid hyperplasia which may occur in the thyroid gland.

At one time some confusion also existed regarding Hashimoto's disease and malignant tumours of the thyroid, but in recent years the diagnostic difficulties appear to have been resolved and the benign character of Hashimoto's disease recognized. Joll (1940) stated that neither necrosis or calcification nor abscess formation occur in Hashimoto's disease, that the whole gland is uniformly enlarged, and that

no serious encroachment on extrathyroid tissues occurs.

Shaw and Smith (1925) described 6 cases under the heading of ' Reidel's chronic thyroiditis ', and in 3 of these some focal ' round-cell ' infiltration and some diffuse fibrosis affected the adjacent tissues. The descriptions regarding the extracapsular involvement are, however, quite clear : they do not suggest a neoplastic permeation and in no way resemble the appearances found in *Case* 1. These authors believed that Hashimoto's disease was an early stage of Reidel's fibrosis and regarded their cases as demonstrating successive phases in the evolution of the disease, but Graham (1931) later doubted whether any of the cases conformed to Reidel's description and considered that 4 of the cases were examples of Hashimoto's disease. However this may be, it is recognized that Reidel's disease and some other types of ' thyroiditis ' may involve extrathyroid tissues, and a distinction from neoplastic infiltration may have to be made at operation and during pathological examination of a thyroid gland.

In our first case there is a distinct change from the histological pattern typical of Hashimoto's disease to that seen in the neoplastic tissue involving the right lobe, the isthmus, and the extrathyroid tissues. There seems no doubt that we are dealing with a malignant neoplasm which shows frequent mitoses, many abnormal types of mitosis, areas of necrosis and a high proportion of cells resembling reticulum cells, and which infiltrates and destroys voluntary muscle. There is no sign of fibrosis in the extrathyroid tissues and the ' chain-like ' permeation of tumour cells in the connective-tissue planes between muscle-fibres is characteristic of neoplastic infiltration and quite unlike a cellular ' inflammatory ' infiltration.

In our second case the opportunity arose to examine the thyroid tissue at two intervals of time.

The first specimen showed the features of Hashimoto's disease. The distribution and the arrangement of the lymphoid tissue were typical. Many of the acini were lined by cells of Askanazy type, a feature described by many authors (Womach, 1944 ; Crile, 1948 ; Lennox, 1948 ; Marshall, Meissner, and Smith, 1948), though some have referred to them as acidophil cells or Hurthle cells. Intra acinar lymphoid cells were present as mentioned by Joll (1939) and Schilling (1945) and in one place there was squamous metaplasia (Marshall and others, 1948). There were some coarse collagen bundles, but no fine fibrosis. The unusual feature was the reticulum-cell proliferation, and it was on this ground that malignancy was first suspected.

In the second specimen undoubted reticulum-cell sarcoma was present, but recognition of Hashimoto's disease was difficult though possible in some parts of the specimen.

The question regarding the true nature of the neoplasms in our two cases can be formulated thus : Are these reticulosarcomata or anaplastic carcinomata ?

This problem, which arises also in many other sites in the body, is an extremely difficult one. In routine histological work one is unconsciously guided by the site of the neoplasm : consciously by the results of a careful search for evidence indicating a transition between the sarcoma-like tissue and unequivocal cancer ; and also to some extent by the pattern of the reticulin network in the tumour.

None of these aids is foolproof, however, and Scott (1952) has recently demonstrated the pitfalls in the diagnosis of these tumours. He concludes that no case should be finally assessed until a complete post-mortem examination has been made. Fortunately for the patients, this has so far been denied us, but points in support of the diagnosis of reticulosarcoma may be enumerated. The cytological features of the neoplasm are identical with reticulosarcoma as found in lymph-nodes and other lymphoid tissues of the body. It is clearly arising in association with the lymphoid hyperplasia in these thyroids affected by Hashimoto's disease. Search of many sections fails to reveal any areas of neoplastic tissue suggestive of a carcinomatous nature. Reticulin stains reveal a network intimately associated with the tumour cells, accompanying them also into the extrathyroid tissues in *Case* 1.

Study of the literature reveals a close relationship between reticulosarcoma and pre-existing lymphoid hyperplasia in the thyroid gland. Such an association has been recorded by Rice (1932), Graham (1931), Vaux (1937), Adé (1941), Kellett and Sutherland (1949), Dinsmore and others (1949), and Lindsay and Dailey (1955). The neoplastic change does not seem to be related to any one particular type of thyroid disease (e.g., Hashimoto's disease, Reidel's thyroiditis), but simply to the presence of lymphoid hyperplasia from whatever cause. Kellett and Sutherland were not able to establish that Hashimoto's disease was the pre-existing condition in their cases since in only three of these was there sufficient tumour-free tissue available for study. In these the lymphoid hyperplasia was not sufficiently widespread and germinal centres were absent, except in one case where they were atypical.

We believe that, in our cases, where most of the thyroid glands were available for study, it is established that a reticulosarcoma has developed in glands affected by Hashimoto's disease.

SUMMARY

Two reticulosarcomas of the thyroid are recorded in a woman and a man, both aged 58 years. These appear to have developed in glands affected by Hashimoto's disease.

Study of the literature indicates that many of these tumours have been associated with pre-existing lymphoid hyperplasia in the thyroid and, although this is due to Hashimoto's disease in our cases, it appears that lymphoid hyperplasia from any cause may be the origin of such tumours.

Our thanks are due to Professor Hadfield, who studied the material from our first case and confirmed the pathological diagnosis. We should also like to thank Dr. G. S. Sansom for the photomicrographs.

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EXCISION OF A CARCINOMA OF THE MIDDLE THIRD OF THE ŒSOPHAGUS WITH AORTIC GRAFT REPLACEMENT—PRELIMINARY REPORT

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THIS paper describes a method of restoring continuity following resection for carcinoma of the œsophagus. Its use has been considered only in juxta-aortic growths where cure by the classical methods has not been possible. The method to be described consists of exploring the lesion in the mid-œsophagus through the right pleural cavity, resecting the growth with adequate margins of normal œsophagus when possible, and restoring continuity by suture of an aortic graft to the upper and lower ends of the divided œsophagus. The graft has been taken, prepared, and preserved in 4 per cent formalin in the manner described by Nuboer (1954). Such experimental evidence as there is suggests that the use of vascular grafts to bridge tubular defects conducting fluid other than blood, results in obliteration of the lumen of the graft by fibrosis. Schein, Sanders, and Hurwitt (1955) used autogenous splenic arterial grafts to bridge defects in the ureter of the dog. They found that after a 90-day period the graft underwent fibrosis with obliteration of the lumen. Because of the possibility of fibrosis, cases of œsophageal carcinoma treatable by any other method were excluded from the series. In this preliminary report the reasons for adopting the technique, a brief description of the operation, and reports on three cases will be presented. From five patients treated by this palliative procedure only one died in hospital—a man of 78 from leakage of the upper suture line on the sixteenth day.

Reasons for Adopting Present Technique.—These derive from (i) the application of a number of observations made during a two-year trial of the method of polythene tube replacement (Berman, 1952 a) for lesions of the mid-œsophagus, and (ii) a study of earlier work on means of restoring continuity of the œsophagus by foreign material combined with the accumulated data from publications on the fate of aortic grafts when used for replacement in the aorta. These two reasons will be discussed more fully.

i. In the two-year trial of the method of resecting the growth and inserting a polythene tube to restore continuity 12 patients were operated on. The results were disappointing. Failure to achieve satisfactory palliation may have been due to the type of tube used and the method of using it, as much as to the practical shortcomings of the procedure. The causes of death were leaks from one or both ends of the tube, lung abscess from inhalation of regurgitated stomach contents up the tube, and pressure by the tube on the heart causing gross disturbances of rhythm, and are reflections of three physical characteristics of this prosthesis which obviously are undesirable. These are the difficulty in achieving a leak-proof union between polythene and œsophagus, the conversion of the œsophagus by the tube into a channel with a permanently open lumen allowing free regurgitation from stomach to pharynx especially when the tube needs to be a long one, and finally the relative rigidity of the tube which presses on the heart instead of moulding itself into the mediastinum as a normal structure free of pressure effect. These last two objections might be remedied by using a softer tube as suggested by Domm, MacManus, Anderson, and McCormick (1955). Our method of securing the tube by placing a purse-string suture round the œsophagus and tying the tube in, led to necrosis of the portion of the œsophagus beyond the purse string, loosening of the suture and displacement of the tube. In two cases this occurred as early as the third post-operative day. Direct suture of œsophagus to the rim of the tube as an end-to-end anastomosis is difficult because of the consistency of the polythene. We should use, on a future occasion, a tube with the rim drill-holed to a size corresponding to the size of thread used for the anastomosis and pass a suture through each of these holes and into the œsophageal wall. The union that is made between the tube and the œsophagus is in most cases only temporary and the tube has usually worked loose, in our experience of the Berman (1952 a) operation. In favourable circumstances a track forms by the tenth day which allows the patient to swallow even after removal of the tube. No convincing evidence is available, at autopsy, that this track

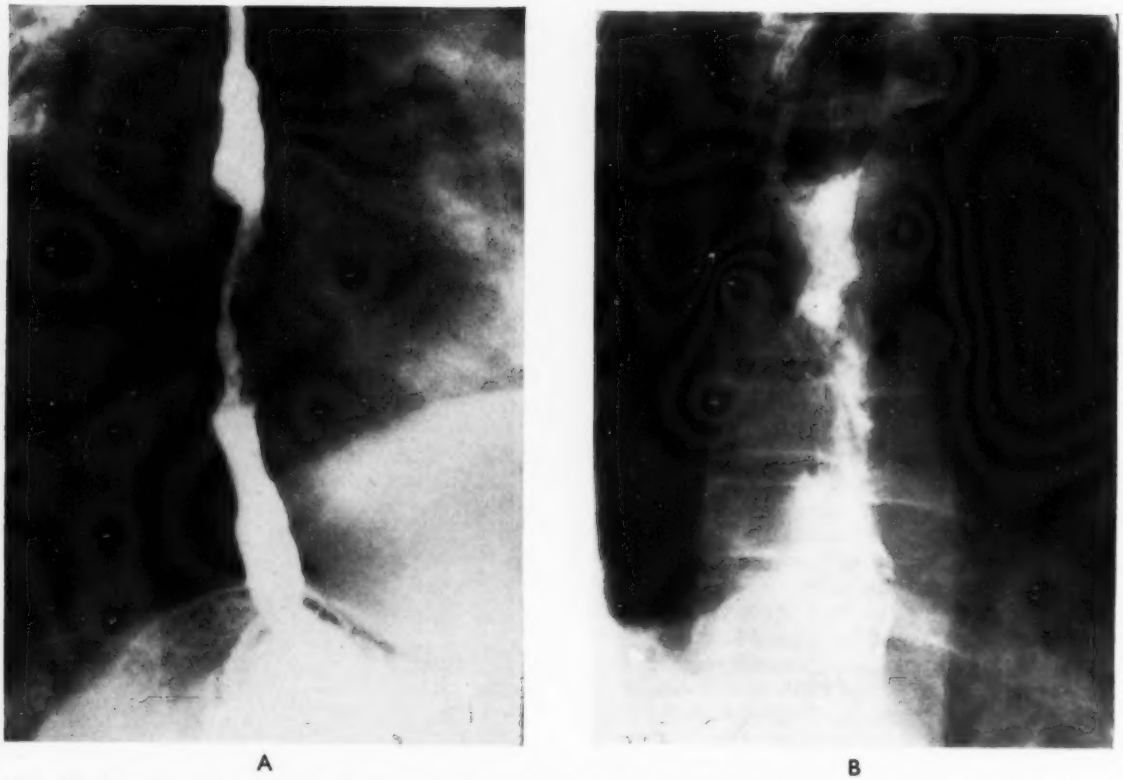
becomes epithelialized. From these observations in this small series of polythene tube replacements an alternative material was sought which could be used to produce a leak-proof union of the œsophageal ends, had none of the disadvantages of a rigid tube, and would serve as a scaffold for the ingrowth of œsophageal epithelium if in fact this did occur. Support was also given to the belief that the œsophagus need not be an entirely muscular tube for deglutition to be a satisfactory sensation for the patient.

ii. Previous publications on unnatural ways of restoring the œsophagus after resection are few compared with descriptions of methods of repairing the trachea. Rob and Bateman (1949) described a case in which continuity of the cervical œsophagus was restored by means of tantalum gauze covered with fascia lata. Klopp, Alford, and Pierpont (1951) used polythene film with split thickness skin grafts for repair of the cervical œsophagus. Berman (1952 b) reported satisfactory results in 13 of 20 dogs in which a segment of œsophagus was replaced by a polythene tube. The results of this procedure in the human are variable. As a means of approximating the ends of the œsophagus an aortic graft seems an obvious choice. Its diameter corresponds closely to that of the œsophagus, and its flexibility and toughness are unaltered in preparation. A more compact combination of qualities, in particular the propensity of the graft to hold sutures, seems unobtainable. A number of publications have described the fate of homologous aortic grafts used for aortic replacement. These suggest that the blood-supply to the graft originates from the surrounding tissue and that the graft itself forms a scaffold for the ingrowth of host tissue (Debakey, Creech, Cooley, and Halpert, 1954; Coleman, Deterling, and Parshley, 1955; Gwathmey and Thompson, 1955). Whilst it is unlikely that this relationship between graft and host tissue is accounted for solely by it being part of a blood-conducting channel, there is no reason for supposing that in an œsophageal medium the aortic graft and host tissue will behave in the same fashion. For this reason and in the absence of any information on the late effects of the procedure only cases in which we were prepared to see a fibrous stenosis replace a malignant stricture have been treated by this method. It has the advantage over other palliative measures that it offers the patient some small chance of cure because the lesion is resected. Our impression is that though this method itself may not be acceptable, some comparable means of providing a channel out of tissue locally, without the support of any foreign material, will evolve for certain high œsophageal cancers which at present are virtually untreatable by open operation. The high operative mortality of current methods is largely that of the means which, from necessity, must be used to restore continuity rather than the mortality of excising the cancer.

The Operation.—An appropriate right rib is resected, the pleura opened, and the azygos vein divided. The lesion in the œsophagus is mobilized leaving the opposite pleura intact whenever possible; if opened it is carefully sutured. This is done to preserve a pleurally covered channel for the graft to pass through and is an important detail. The growth with adequate margins of normal tissue is resected with any glands that can be reached. Clamps are not placed on the œsophageal ends which are allowed to retract. The graft is measured against the length of the deficiency with the ends of the œsophagus in the retracted position. Any shortening of the graft that needs doing is taken from the end of lesser diameter. This end is then sutured to the lower end of the œsophagus by about a dozen sutures of

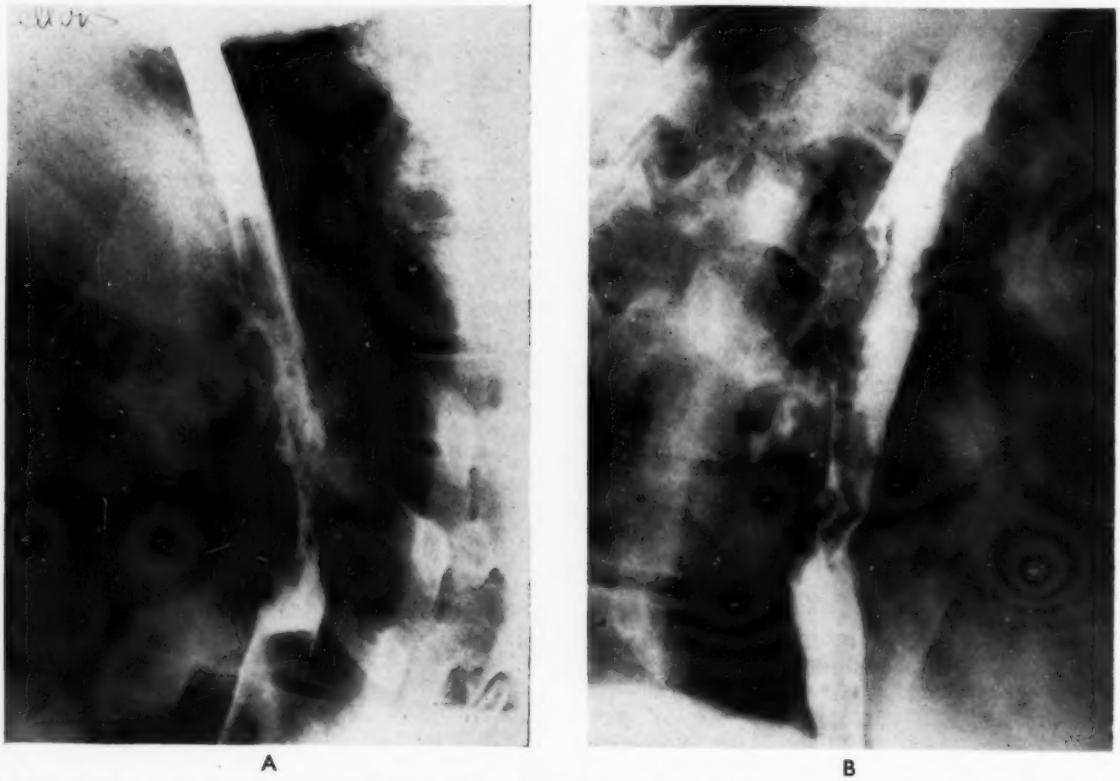
FIG. 692.—*Case* 1. Post-operative barium swallow. The upper suture line can be seen. Some barium passes outside the aortic graft.

No. 40 thread, placed interrupted fashion equally around the circumference with the knots tied on the inside. The lower end is anastomosed first. The bed in which the graft is to lie is made hæmostatic; any bleeding raises the covering pleura off the graft, and as so much seems to depend on this covering absolute hæmostasis is essential. The upper end of the graft is trimmed to an exact length and suture of the graft to the upper end of the œsophagus carried out in the same fashion as at the other end. The graft is shut off from the main pleural cavity by approximating the mediastinal pleura or, if this has been taken, by utilizing pleura turned back off the chest wall. Pleura is sewn to the graft at both ends of the anastomosis, reinforced by the azygos vein at the upper end and the pleura of the pulmonary ligament at the lower. A number of fine thread sutures approximate the pleura over the graft along its length, and the aim has been to make this a barrier against spread of infection and support to the graft by as complete and strong a closure as possible. It is advisable to confirm that no collection of blood has lifted this pleural covering off the graft before closing the chest. The pleural cavity is drained only if there is an air leak from the lung. The operation



A B

FIG. 693.—*Case* 2. A, Pre-operative barium swallow outlining the neoplasm. B, Post-operative swallow (postero-anterior view). Screening shows an apparently normal œsophagus.



A B

FIG. 694.—*Case* 3. A, Pre-operative swallow showing irregular narrowing of the œsophagus. B, Post-operative swallow. Screening shows an apparently normal œsophagus.

should take no longer than one-and-a-half hours and the convalescence is remarkably easy and free from distress.

The operation described is the method used. It is the result of certain technical details revealed during our trial of the polythene tube replacement operation (Berman, 1952 a), rather than a method which we have applied only to those cases in which an aortic graft has been used. Orr (1956) has performed a similar operation on two occasions using an aortic graft, with satisfactory immediate results.

CASE REPORTS

Case 1. W. W., a boilerman, aged 58, consulted his doctor because of increasing dysphagia over four months. When first seen he was in poor general condition and was unable to swallow either solids or liquids. A preliminary gastrostomy was done for feeding purposes. The barium swallow showed an obstructive lesion at the level of the tracheal bifurcation. Œsophagoscopy demonstrated a hard irregular mass obstructing the œsophagus at 28 cm. from the incisor teeth ; no biopsy was taken. At bronchoscopy some narrowing of the left main bronchus was present although actual involvement of the bronchial wall by growth could not be detected. On account of the local findings and his poor general condition no radical operation was contemplated. The patient was in considerable distress. Four weeks after the gastrostomy the œsophagus was explored. At operation a large tumour some 10 cm. in length was found centred beneath the arch of the azygos vein ; it extended and was fixed to the aortic arch by direct extension. After resection of the involved segment a portion remained on the aorta. (Examination showed the lesion to be an epidermoid carcinoma.) A 15-cm. aortic graft taken from a 28-year-old male five weeks previously was used to restore continuity and inserted by the method already described. The diameter of the graft seemed unusually large. The patient was fed entirely through the gastrostomy for ten days and made a satisfactory convalescence (a small contralateral effusion was aspirated on the tenth day). He started on fluids on the eleventh post-operative day and by the twentieth day was able to eat the meal he had been wanting for four months—bacon and eggs. The barium swallow on the twenty-fourth day is shown (*Fig.* 692). The patient continued to swallow normally for the next four months when a recurrence of the growth obstructed the graft. The patient died from bronchopneumonia two weeks later. No autopsy was obtained.

Case 2. E. B., aged 47, a frail lady weighing 7 stone, with a blood-pressure of 250/140 mm. Hg, had a history of four months' increasing dysphagia for solid food. A barium swallow showed a stricture of the œsophagus extending downwards from the level of the tracheal bifurcation for 4 cm. (*Fig.* 693 A). Bronchoscopy was normal and œsophagoscopy yielded a biopsy showing a well differentiated squamous carcinoma 29 cm. from the incisor teeth. Her general condition precluded a radical operation. Because it was proposed to keep the patient without fluid by mouth for ten days a preliminary gastrostomy was done. Two weeks later an 8 cm. segment of the mid-œsophagus was removed ; the removal was apparently complete but microscopy later showed malignant cells present in the upper free margin. A 10-cm. graft, taken from a 24-year-old male seven weeks previously, was inserted. The pleural cavity was drained. Convalescence was satisfactory, the patient swallowing normally from the tenth day. Barium swallow at three weeks showed a normal passage down the graft (*Fig.* 693 B). Four weeks after the operation the patient was œsophagoscoped ; what was thought to be a tangled mass of hair was seen at 28 cm. from incisor teeth. One of these was grasped by biopsy forceps and gently withdrawn ; it was found to be a suture and with it the whole graft was removed. The graft obviously had not been incorporated locally. Apart from being a little more limp than at the time of its insertion one month previously, its general texture was unaltered. It readily held sutures at either end, and its length was unchanged. The original sutures were present on the graft and had cut out from the ends of the œsophagus. Microscopy (Dr. A. P. Prior) was as follows : " An inflammatory reaction affects both the media and the adventitia of the graft. This tends to be most marked at the ends of the specimen, but there is no evidence of epithelialization. Also at the ends there is penetration of the graft by inflammatory cells for a few millimetres. Sometimes there are collections of red cells between the fibres of the graft suggesting the possibility of revascularization of the graft itself. The collagenous tissue is largely preserved but there appears to have been destruction or absorption of large parts of the elastic tissue." The patient continued to swallow, but ten weeks after operation developed recurrence of growth in the region of the upper suture line. A Souttar's tube was inserted.

Case 3. N. F., aged 47, a lorry driver, complained of two months' increasing dysphagia and painful swelling of the hands and ankles. When first seen he was unable to take anything solid and had considerable swelling of both hands with marked finger clubbing which had appeared recently. He had a productive cough. A radiograph of the chest was normal. A barium swallow (*Fig.* 694 A) demonstrated a filling defect in the upper œsophagus. Bronchoscopy showed a bulge in the posterior wall of the trachea about 4 cm. above the carina, and œsophagoscopy an obstructive lesion of the œsophagus 23 cm. from the incisor teeth. A biopsy from this area was reported as an epidermoid carcinoma. The local findings indicated that only a palliative operation was possible. At operation a growth was found adherent to the posterior wall of the trachea. Complete separation from the trachea was not possible. The growth was resected and the œsophageal ends joined by a 14-cm. graft taken from a 23-year-old male six weeks previously. A gastric tube was passed from the mouth through the graft into the stomach through which the patient was fed for 48 hours. Twelve days after operation he had what he described as his best meal for three months. A barium swallow sixteen days after operation showed a normal passage down the œsophagus (*Fig.* 694 B). He was relieved of the swelling of his hands and ankles within 48 hours of operation and continues to swallow normally.

We should like to thank Dr. A. P. Prior for procuring the aortic grafts and to Dr. M. Israelski for the radiographs and reports on screening.

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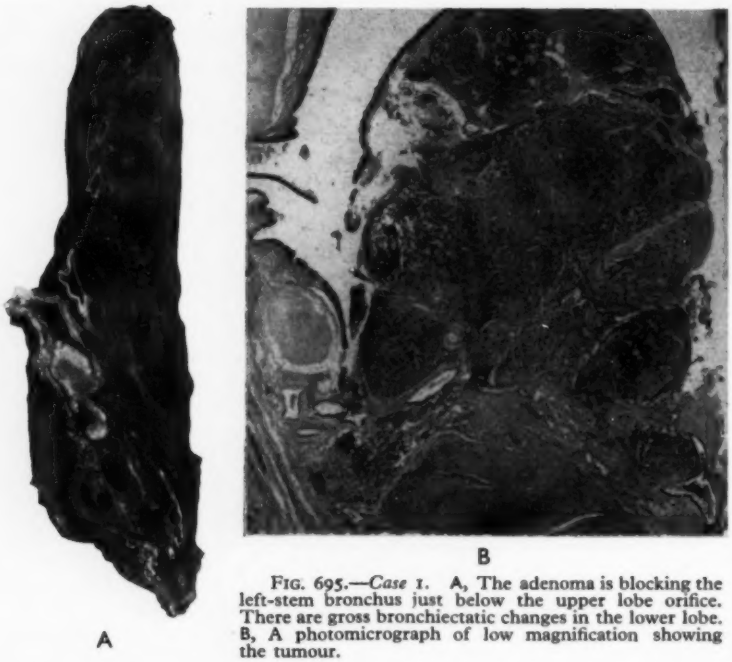
BRONCHIAL ADENOMA

By KENNETH FRASER, Glasgow

Formerly bronchial adenoma was looked on as something of a rarity, but now these tumours are being found more frequently following the wider use of bronchoscopic examination in the investigation of chest disease. In the past thirteen years there have been 12 cases in the surgical thoracic unit of Hairmyres Hospital. Nine of the cases here described were briefly referred to by Dick (1950) in a Honeyman Gillespie Lecture.

At one time it was sufficient to consider bronchial adenoma as a tumour having little variation from case to case. Reports in more recent years have underlined the fact that these tumours occur in at least two forms, the carcinoid and the cylindroma variants of bronchial adenoma. Carlens, Wiklund, and Bergstrand (1954) believe that these two are variants of the same kind of tumour and that transitions between the two types can commonly be seen in one case.



A B

Fig. 695.—*Case* 1. A, The adenoma is blocking the left-stem bronchus just below the upper lobe orifice. There are gross bronchiectatic changes in the lower lobe. B, A photomicrograph of low magnification showing the tumour.

The 12 cases here reported are all of the carcinoid type of adenoma.

HISTORICAL

Bronchial adenoma was first described in post-mortem material in 1882 by Mueller. According to Goldman and Conner (1950) the condition continued to be diagnosed post mortem only until 1930; in that year the bronchoscopic recognition and bronchoscopic treatment during life commenced with the correct diagnosis by Kramer (1930). This phase remained as the only approach to the problem and its treatment until 1938, when Hamperl introduced the more radical and much more conclusive method of treatment by pulmonary resection, in conformity with the more complete understanding of the lung damage which results in many of these cases from bronchial blockage.

INCIDENCE AND AETIOLOGY

Sanders and Kingsley (1948) found the incidence of adenoma of the bronchus at the Lahey Clinic to be 6·9 per cent of all proved bronchial tumours.

The origin of these tumours favoured by most authorities is the epithelium of the mucous glands and ducts of the bronchi (Brann and Goldman, 1941). It would seem that whereas in bronchial carcinoma the usual site of origin is in the bronchial epithelium, in adenoma this is never the cell of origin, and in this way adenoma resembles fibro-adenoma of the breast, having origin from the acini while carcinoma arises from the duct epithelium. Carcinoma may arise, and often does, in smaller bronchi, adenoma seldom if ever takes origin distal to the main lobar bronchi.

This latter feature could be used as an argument in favour of the view of origin suggested by Womach and Graham (1938). They put forward the very reasonable view that adenoma arises as the result of failure of a bronchial bud to develop into the orderly arrangement of tissue found in normal lung; any such tissue will obviously be associated with, and remain intimately connected to, the stem bronchus. When mesodermal elements preponderate they believe that chondroma, lipoma, and fibroma will occur, and when entodermal elements are in the majority that an adenoma will result. In support of this view they hold that adenoma is difficult or impossible to differentiate from fœtal lung. In further evidence they have shown that other anomalies may be present in these cases, including absence of lobulation or excessive lobulation and congenital cystic disease. When the author visited Dr. Graham at his clinic at the Barnes Hospital, St. Louis, in September, 1951, he was still in favour of this view and he showed specimens of 2 cases in which there was a carcinoma and a sarcoma growing side by side; this was held to support the theory of failure in the development of part of a lung. His explanation of this rare association in the realm of tumours was that it had occurred where a bronchial bud had failed to develop and the mesodermal and entodermal elements had undergone simultaneous malignant growth.

Mallory (1946) believes that adenomata are similar to carcinoid tumours of the appendix, having slowly growing cells with affinity for silver stains. Several of the cases described in this article were studied by H. E. Hutchison and the author at the Western Infirmary, Glasgow, in an attempt to determine whether the general histological resemblance

to the carcinoid tumours was accompanied by an argentaffin reaction and other histochemical similarities. In no case could we confirm Mallory's findings and we therefore concluded that the resemblance was only superficial.

PATHOLOGY

Bronchial adenoma is a tumour of slow growth, almost invariably arising in relation to a stem bronchus of a lobe or lung; consequently it was seen on bronchoscopic examination in every one of the 11 cases bronchoscoped. Moersch and McDonald (1950) reported that the tumour was seen in 91 per cent in a series of 84 patients at the Mayo Clinic.

The gross characteristics of these tumours are so typical that the bronchoscopist at the time of examination can often suggest the diagnosis of adenoma. The picture seen is the partial or complete occlusion of a stem bronchus to a lung or lobe by a lobulated pink, red, or purplish mass which is firm to touch and bleeds readily. The surface of the tumour is smooth because as a rule the bronchial mucosa is intact over the tumour and has not ulcerated, though ulceration may develop and squamous metaplasia may occur. An important feature not fully appreciated in the early days of treatment was that some of these tumours resemble an iceberg floating in water, in that the visible portion is much smaller than the portion lying within the lung substance, but outside the bronchus. While it is true that some have no extrabronchial growth (*Cases* 1, 2, 3; *Figs.* 695, 696), most adenomas show some extrabronchial extension. Such tumours, with no extrabronchial extension, will probably have a narrow pedicle and so be freely movable and thus be readily excised through the bronchoscope unless they have reached a large size. The more usual tumour might be likened to a dumb-bell. It has a small intrabronchial portion joined by a narrow neck of tumour which passes through the cartilaginous rings to the main mass lying in adjacent lung tissue, which is compressed around it (*Case* 4, *Fig.* 698). It is therefore quite impossible in these cases for the bronchoscopist to tell how large the tumour really is.

If the adenoma partly or completely blocks the bronchus into which it projects, the lung distal to the obstruction will show pathological changes of varying severity. Such obstruction may cause segmental or lobar atelectasis (*Cases* 2, 5, 6; *Figs.* 696, 699, 701). Subsequent stagnation of the secretion will lead to superadded infection and will give rise as a common sequel to bronchiectasis (*Case* 1, *Fig.* 695) and lung abscess. McBurney, Clagett, and McDonald (1952) report significant to severe degrees

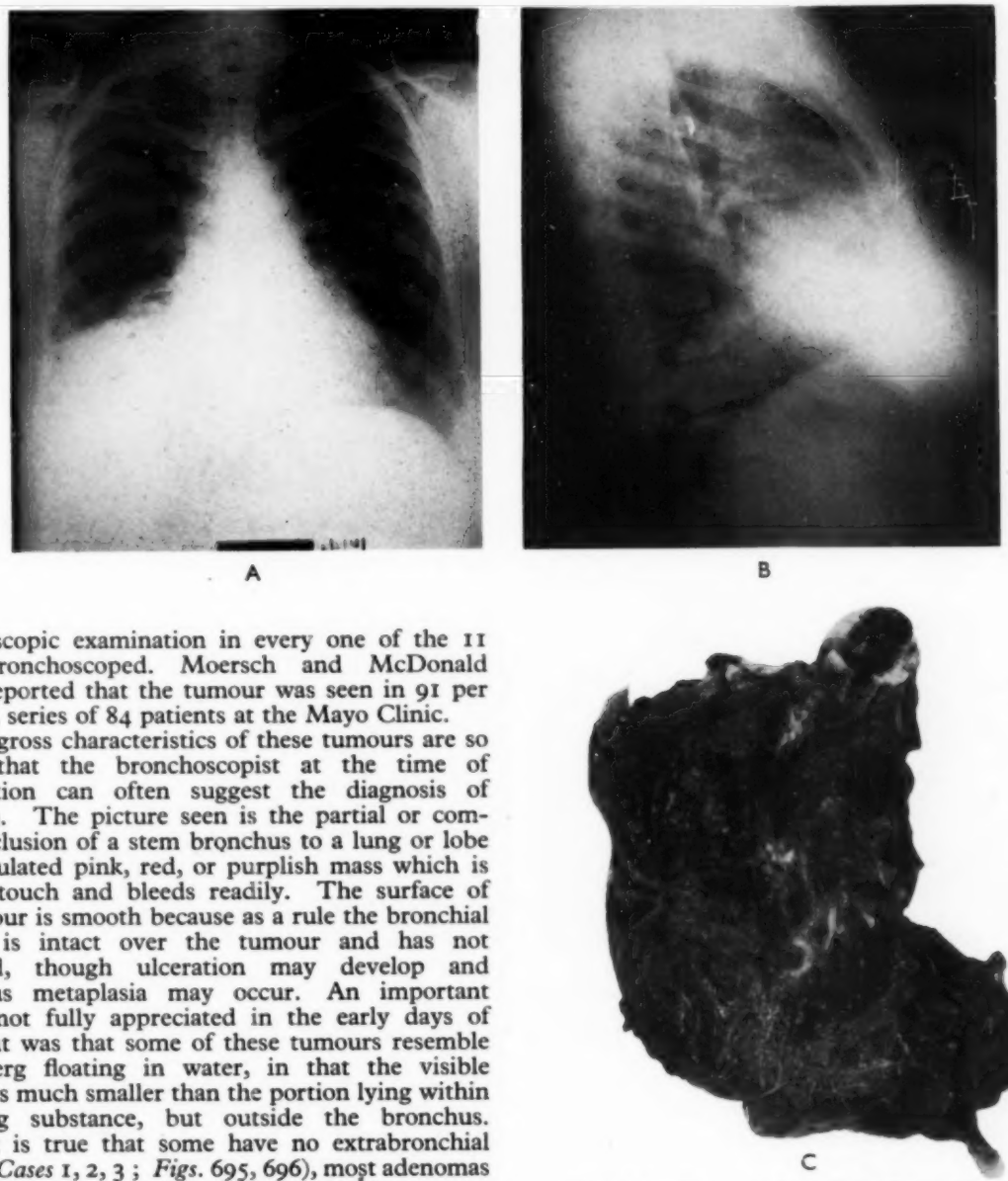
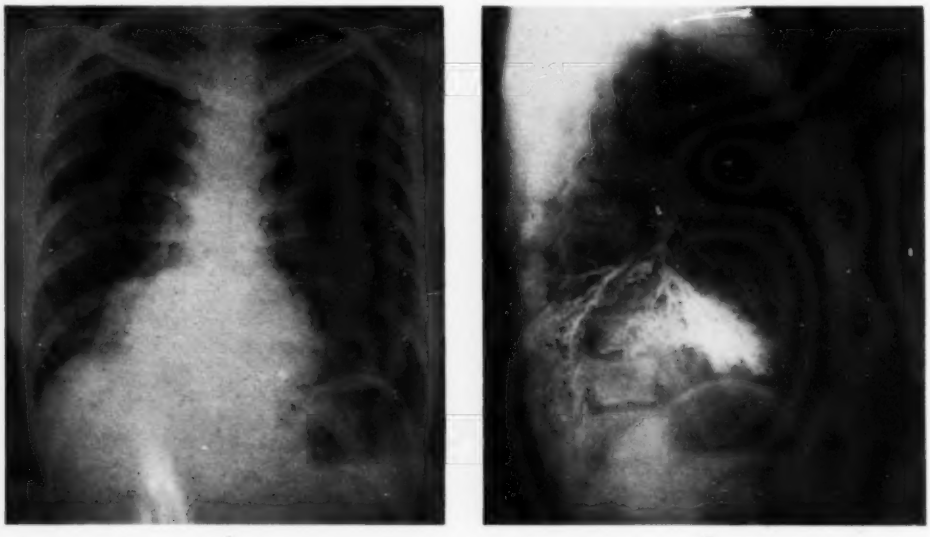


FIG. 696.—*Case* 2. A, B, There is collapse of the right lower and middle lobes due to an adenoma in the right-stem bronchus. C, The tumour was entirely intrabronchial and can be seen projecting from the bronchus at the top right-hand corner of the specimen.

of obstructive pulmonary changes in 58 per cent of 102 cases treated at the Mayo Clinic.

A feature I have not found published in other series of cases occurred in *Case* 7 (*Figs.* 702, 703). On the superior aspect of the lung there was a giant tension cyst measuring 12 × 10 cm. I found in the cyst three bronchial openings which had communication with the upper-lobe bronchus. The cyst lining was smooth, shiny, and pale blue in colour. It is interesting to report that neither the and McDonald (1950) had 3 instances in a series of 86.

Adenoma seen through the bronchoscope shows up as a pink or red lobulated tumour, but when the tumour is sectioned after removal it is usually greyish white in colour and its lobulated appearance is maintained by strands of fibrous tissue running throughout its substance. The largest tumour occurred in *Case* 4 (*Fig.* 698) and was 6 × 4·5 cm. As might be expected, this was the case giving the longest



A B

FIG. 697.—*Case* 4. A, B, The tumour is blocking the middle segment of the right lower lobe. The spherical shadow is due to the tumour. Tne lipiodol is held up at the origin of the middle segment bronchus.

clinicians nor the radiologist diagnosed the condition as a cyst; all thought there was a pneumothorax with a concomitant effusion. For five years she had been frequently admitted to a sanatorium as a case of tuberculous effusion, but this was never proved; indeed, not only was the fluid tapped on several occasions and found to be serous in character, but attempts were made to re-expand the collapsed lung by withdrawing air from the 'cyst', as much as 3000 c.c. of air being removed on one occasion. As one would expect in view of the bronchial communications, this had no effect. This case is a good illustration of the necessity of having a lateral radiograph in all cases of this kind.

In the 12 proved cases of adenoma, 7 were in the right lung and 5 were in the left. This preponderance of right lung over left is a frequent feature in published series. Moersch and McDonald (1950) had 47 right tumours as against 38 left ones. Five of the 12 specimens were in the stem bronchus, 4 were in the bronchus to lower lobe, 2 in the upper lobe, and 1 in the middle lobe.

Although Baldry (1952) reported 9 cases of carcinoma of the middle-lobe bronchus from a series of 185 (5 per cent), this figure is somewhat higher than other reports and it is generally agreed that carcinoma of the middle lobe is uncommon. On the other hand, one has the impression from the literature that adenoma of the middle-lobe bronchus occurs rather more frequently than does carcinoma. *Case* 5 (*Fig.* 700) occurred in this site. Moersch history, namely 26 years. This tumour appeared to be invading bronchus at one point and this may have accounted for the marked increase in symptoms shortly before admission to hospital. These included three large hæmoptyses, following one of which the patient became unconscious. A further interesting feature of this case was the presence of a small grape-sized congenital cyst in the lower edge of the lobe.

The specimens removed from those cases in which the tumour projected into the lung substance showed that tumour growth had displaced and compressed the lung tissue before it in such a way as to ensheath the tumour and give the appearance of a false capsule (*Cases* 4, 5, 8; *Figs.* 698, 700, 705). *Case* 5 (*Fig.* 700), in which the tumour was in the middle lobe, illustrates these features particularly well. The tumour had been seen on radiography 10 years previously but no noticeable change in size had occurred in spite of recurring attacks of hæmoptysis every 3 or 4 months for the first 8 of these years. At bronchoscopy the tumour was seen to project into the stem bronchus from the middle-lobe orifice and it was partly obstructing the main lower-lobe bronchus. *Fig.* 700 illustrates this projection of the tumour; indeed, the projection was so marked that at operation it was found necessary to remove the lower lobe as well as the middle lobe, and some difficulty was encountered in leaving sufficient stem bronchus behind so that the closure would not encroach on the upper-lobe orifice.

Although bronchography showed no bronchiectasis in the lower lobe it will be seen from the photograph of the specimen that there were present early bronchiectatic changes (*Fig.* 700). The tumour had reached such a size that there was only a thin layer of the middle lobe left surrounding it. This case is a good illustration of the impossibility of completely removing some tumours by bronchoscopy; even if it had been possible to do so only grossly diseased lung tissue would have been left behind, tissue which could never have regained any useful function.

In *Cases* 1, 2, 3, 6, 9, and 10 the tumours were examples of intrabronchial adenoma having little if any extrabronchial extension. Of these, 3 had narrow pedicles of attachment to the bronchial wall; in 1 (*Case* 9, *Fig.* 706) it was possible to remove the tumour through the bronchoscope, and 1 was removed by bronchotomy. All of these cases illustrate the damage that accrues from partial or complete blockage of the bronchus by the tumour, and all showed varying degrees of bronchiectasis, the most marked being *Case* 1 (*Fig.* 695). The changes were sufficiently marked in all except *Case* 3 to require the removal of the affected lung tissue by lobectomy or pneumonectomy. The 2 polypoidal examples with broad bases of attachment to the bronchial wall were both subjects of errors in diagnosis. *Case* 1 (*Fig.* 695) was incorrectly diagnosed as an oat-celled carcinoma from the tissue removed at bronchoscopy, but correctly diagnosed from the post-operative tissue; *Case* 10 (*Fig.* 707) was originally diagnosed as an oat-celled carcinoma until 3½ years later doubts were raised by the receipt of a report from Perth Royal Infirmary stating that the patient was well. On reviewing the specimen and the histology it was found that the tumour had been an adenoma. *Fig.* 707, showing the gross specimen, should clearly raise doubts in the expert's mind that this could have been a carcinoma, the polypoidal nature of the tumour and the absence of invasion beyond the bronchus wall being quite contrary to the usual appearance of carcinoma. Had further evidence been required it was to be found in the history of breathlessness for 20 years accompanied by a ticklish cough for the last 6 years of this period.

HISTOLOGY

The common histological picture shows closely packed cuboidal or columnar cells one or more layers in depth which in some cases may surround a central space of varying size. The cell nuclei are round or oval, and either show dense nuclear chromatin or are vesicular and have small dense nucleoli; characteristic of each specimen was the uniformity of the cells (*Case* 5, *Fig.* 700). Mitotic figures are rarely seen. The pattern in the tumours presented here is of a more solid nature, frequently with solid acini. In others the cells may be grouped more erratically in sheets, cords, or columns with little, if any, tendency to the formation of intercellular spaces, but the arrangement is still regular in that there is no evidence of 'unruly' growth (*Case* 5, *Fig.* 700). The stroma may be abundant

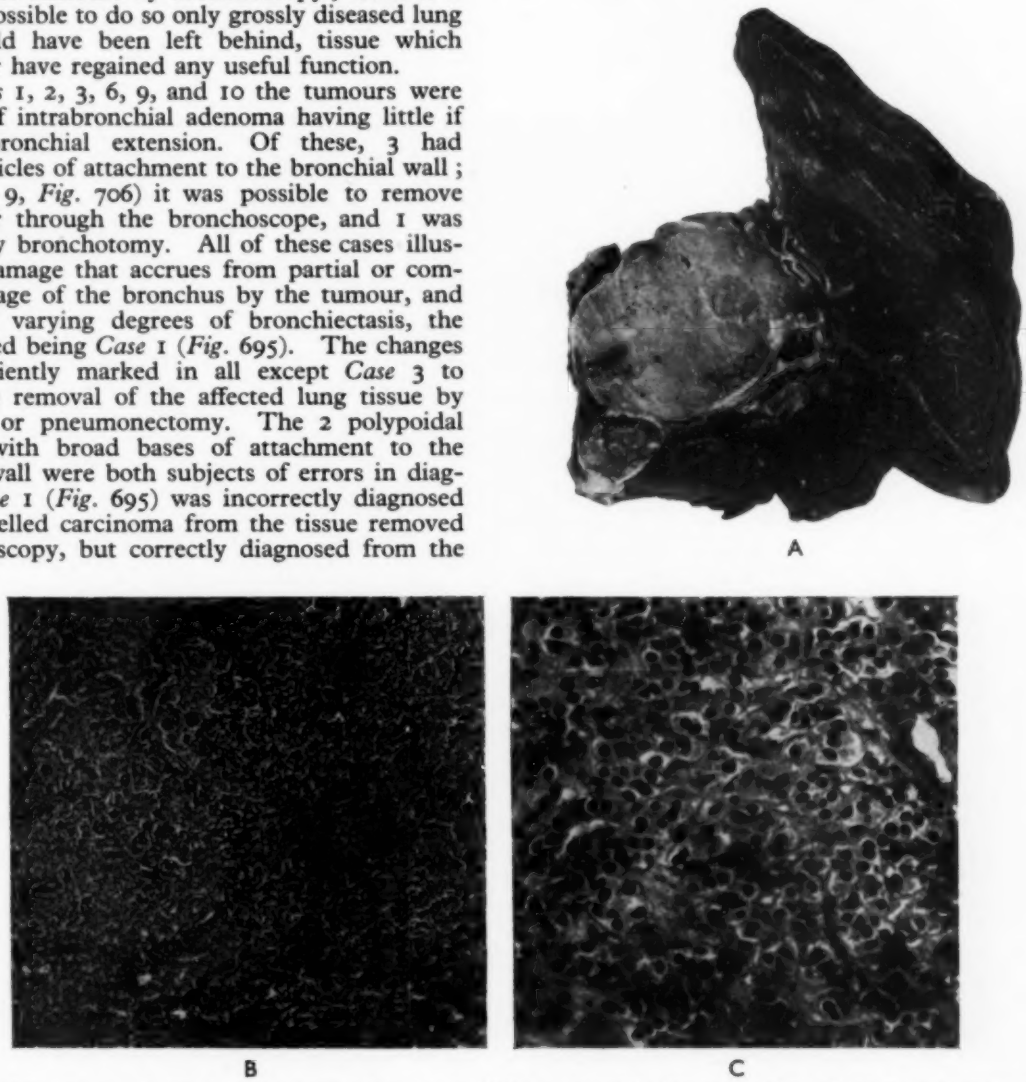


FIG. 698.—*Case* 4. A, The tumour measured 6 × 4·5 cm. It has compressed adjacent lung tissue giving the appearance of a false capsule. There is a small congenital cyst just below the adenoma. B, C, The photomicrographs show abundant intercellular stroma surrounding cells the nuclei of which are deeply stained.

and is richly supplied with capillaries (*Case* 4, *Fig.* 698). The tumour is covered by the normal ciliated epithelium of the bronchus and is separated from it by a layer of connective tissue (*Case* 9, *Fig.* 706). Sometimes ulceration of this epithelium occurs with consequent interruption of this lining, and if the infection persists squamous metaplasia may occur. There was no evidence of metastases to lymph-nodes or to other sites. The only feature of danger occurred in *Cases* 4 and 6 and was looked on as showing commencing invasion in a tumour of long standing. In one the tumour was seen reaching up to a bronchus, the basement membrane of which was ruptured; in the other the tumour was eroding the bronchial cartilage. Several cases have been reported in the literature in which malignant invasion of a bronchus has occurred and in some there has been invasion of lymph-glands. These examples usually occur in the cylindroma type of bronchial adenoma. Although none of the 12 cases here reported was of this group, Carlens and others (1954) found 9 of this type as against 53 of the more uniform so-called carcinoid adenoma. In the cylindroma group the cells form regular, rounded clusters; these cells show considerably more chromatin than the carcinoid type of tumour and they are grouped around mucoid-filled central spaces, the mucin of which stains with toluidine blue. These tumours have a predilection for the trachea and stem bronchi. Rarely, a tumour may show the histological picture of both carcinoid type and the Swiss-cheese appearance of the cylindroma group side by side (Carlens and others, 1954).

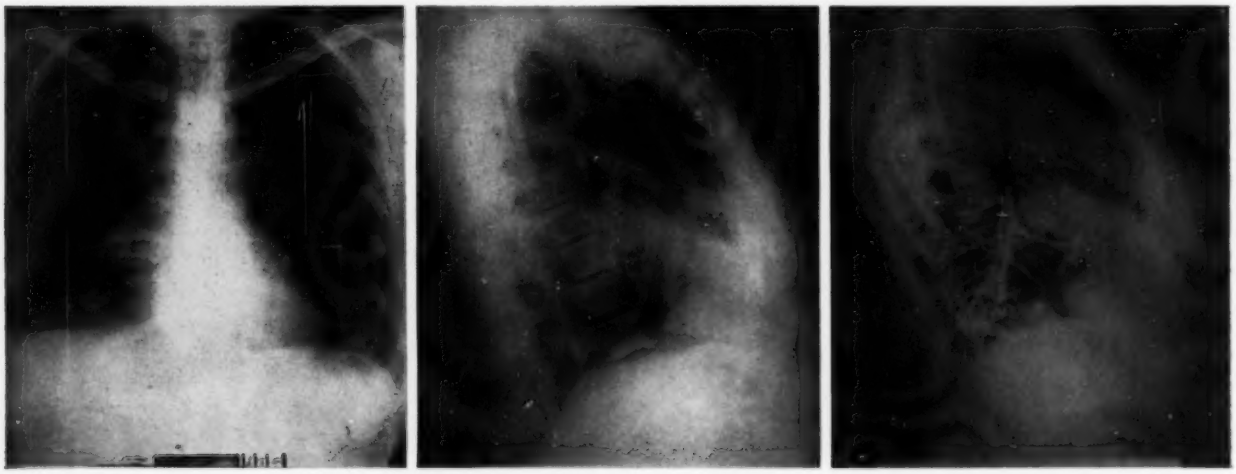


FIG. 699.—*Case* 5. The lateral radiograph shows a triangular shadow adjacent to but in front of the stem bronchus; this shadow is the adenoma which has caused atelectasis of the middle lobe. The lipiodol fails to enter the middle lobe.

CLINICAL ASPECTS

Unlike carcinoma of the lung, adenoma occurs much more frequently in women than in men, the incidence in this series being 10 : 2. The age distribution in adenoma is usually below 40 years and often between 20 and 30. Sanders and Kingsley (1948) reported a case in a child aged 13. In carcinoma, on the contrary, it is most commonly over 40. The age incidence here is higher than usual, the average age being 44, and the oldest patient a male of 67. A further distinguishing feature from carcinoma is the unusually long duration of symptoms along with the maintenance of comparatively good health (*Case* 4). Carlens and others (1954) found on the average that 4 years elapsed before a definite diagnosis was verified in their series of 70 cases.

Because the adenoma projects into a moderate to large size bronchus, cough is the most frequent symptom. It may be dry and unproductive or be accompanied by frothy mucoid sputum. Hæmoptysis in recurring episodes is nearly as common a symptom as cough. It may occur as periodic simple streaking of the sputum or it may be quite profuse in character and sufficiently exsanguinating to cause unconsciousness (*Case* 4). Hæmoptysis was present in 7 of the cases. It occurs at varying intervals and quite frequently months and even years may elpase between the attacks.

Other symptoms associated with adenoma arise as a result of obstruction to the bronchus in which the tumour lies. Thus complete occlusion of the bronchus will lead to atelectasis of a segment or of the entire lobe, and this gives rise to dyspnœa varying in proportion to the amount of lung tissue involved. The radiographs of *Case* 4 (*Fig.* 697) show segmental collapse involving the middle segment of the right lower lobe, while *Cases* 2 and 5 (*Figs.* 696, 699) are examples respectively of lower- and middle-lobe collapse. *Case* 11 (*Fig.* 708) shows complete collapse of one lung. Atelectasis and bronchial obstruction may cause pain in the chest and if infection is superadded fever will occur, and the finding on examination of consolidation leads to an erroneous diagnosis of pneumonia; if this incident is repeated the case is then thought to be one of recurring pneumonia. In time the atelectatic lobe may become grossly infected and all the symptoms of bronchiectasis are superadded.

Radiography may show a shadow, usually near the root of one lung with, distal to the shadow, atelectasis of a segment or lobe of the lung (*Case* 5, *Fig.* 699). This shadow is usually circumscribed and well defined, but it may be partially or completely obscured by the area of atelectatic lung (*Case*

4, *Fig.* 697). Lipiodol instillations may be of help in outlining a blocked segment of the bronchial tree. This is particularly well seen in *Cases* 4, 5, and 7 (*Figs.* 697, 699, 702).

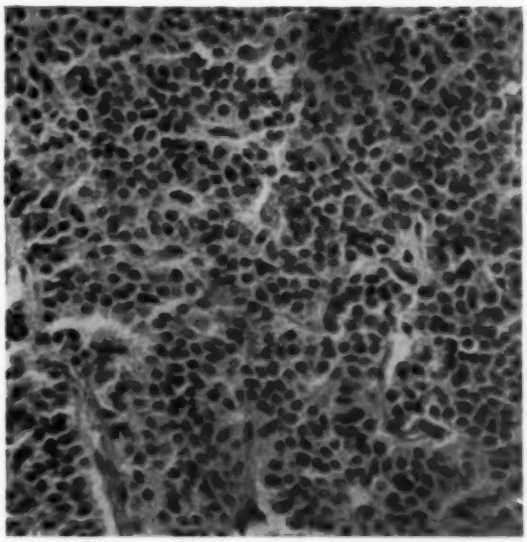


FIG. 700.—*Case* 5. The tumour is seen projecting into the stem bronchus from the middle lobe. The cut specimen reveals the tumour completely replacing the middle lobe. There are early bronchiectatic changes in the lower lobe. The photomicrograph shows typical uniformity of cell pattern.

Final and conclusive diagnosis can be made only on bronchoscopy when the tumour is seen. If the tumour has a narrow pedicle it will be freely movable. When biopsy is taken the tumour cuts crisply and may bleed freely owing to its abundant vascular supply. Biopsy tissue taken at bronchoscopy requires interpretation by an experienced pathologist, for not only is the tissue minute, but the cells may be distorted into an uneven and irregular appearance by the pressure of the biopsy forceps, giving a picture resembling oat-cell carcinoma rather than adenoma. Indeed, in 3 of the cases this was the diagnosis given from the bronchoscopic tissue; in *Case* 9 a further pre-operative bronchoscopy established the correct diagnosis; in *Case* 1 the diagnosis was corrected on examination of the post-operative specimen; but in *Case* 10 it was not until three years after pneumonectomy that a further review of the histology proved it to be an adenoma. In the other 8 cases bronchoscoped the correct diagnosis was reached. In 86 consecutive cases at the Mayo Clinic, Moersch and McDonald (1950) saw

the adenoma on bronchoscopy in 78, and in these a positive biopsy was obtained in 96 per cent. In view of the difficult interpretation of the histology it is as well to warn the pathologist that an adenoma is suspected.

DIFFERENTIAL DIAGNOSIS

The condition most likely to cause confusion is bronchial carcinoma. The long history without marked deterioration of the patient's general health, the comparatively early age at which adenoma usually occurs, and the characteristic bronchoscopic findings should all favour the diagnosis of adenoma.

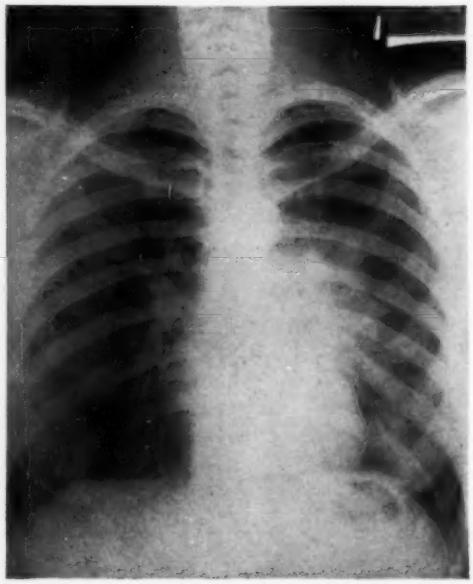


FIG. 701.—*Case* 6. This X-ray appearance resulted from blockage of the left upper lobe by an adenoma. The upper lobe was completely atelectatic.

In the differential diagnosis one has to consider such conditions as 'recurring' or 'unresolved' pneumonia, or a lung abscess, as yet unruptured into a bronchus giving on radiography a solid spherical shadow, usually, however, more peripherally situated than an adenoma. In short, the conditions are very similar to those requiring consideration in the differential diagnosis of carcinoma.

TREATMENT

All 12 cases were treated surgically. One was successfully removed through the bronchoscope, but subsequently required lobectomy to remove diseased lung tissue, while 1 was removed by bronchotomy. Three others were adequately dealt with by lobectomy and 2 required removal of both middle and lower lobes. The remaining 5 cases required pneumonectomy, but in 2 an erroneous diagnosis of oat-cell carcinoma had been made. Lobectomy might have been sufficient in the light of the correct diagnosis and in reference to the specimens (*Cases* 1, 10, *Figs.* 695, 707). All these cases are alive and well; the longest survival is 14 years and refers to the oldest patient, a woman who was 59 years of age at the time of the operation (*Case* 1).

The diagnosis apart, pre-operative bronchoscopy is of great value. It finds the exact location of the tumour, and if a large part of the obstructing tumour is removed drainage from the obstructed lobe will be established and any acute pulmonary suppuration will subside before operation.

The line of treatment adopted will depend on the age and general condition of the patient. Radical treatment by surgery is the generally accepted method of choice, but in an elderly patient or in one of poor general condition all that may be attempted may be simple removal by bronchoscopy of the intrabronchial projection of the tumour. In a healthy patient this method of treatment would seem to be permissible only rarely, where the adenoma is entirely intrabronchial, has a narrow pedicle, and there is no evidence of distal bronchiectasis. If this method is adopted the patient must thereafter be bronchoscoped at regular intervals for years. Initially, a repeat bronchoscopy would be carried out in 1 month's time, and if no recurrence was seen, at increasing intervals up to 6 months or 1 year. If this precaution is not taken and the undeniable risk of recurrence is ignored, the patient will be in danger of developing unsuspectingly an ever enlarging tumour which may well lead to the development of atelectasis and bronchiectasis, or may undergo malignant invasion. The necessity for these repeated examinations is well illustrated by a case of Clagett and Payne (1946). A man of 35 years had an adenoma removed endoscopically from the right lower-lobe bronchus. Two years later he reported well; he was radiographed and no recurrence was seen. A further 3 years elapsed, when he reported with a cough and sputum, and recurrence of the tumour was found. He refused lobectomy and the tumour was again removed through the bronchoscope and radon seeds were implanted. He remained well for a further 5 years when the cough recurred and he developed hæmoptysis. He was again bronchoscoped, and the right-stem bronchus was found to be obstructed by tumour tissue and there was bronchiectasis of the middle and lower lobes. On this occasion the right, middle, and lower lobes were removed and showed marked bronchiectasis, pneumonitis, and fibrosis. The adenoma was $3 \times 2{\cdot}5 \times 2$ cm. and almost entirely extrabronchial.

In the vast majority of cases more radical surgical measures are required. As the obstructing factor will have been present in greater or less degree for many years, the bronchiectasis usually present is unlikely to be reversible; indeed, many of the lobes removed have shown areas of well-marked fibrosis which would render impossible the re-establishment of useful function. As a result it is nearly always desirable to remove the adenoma and the affected area of lung distal to the tumour. Usually this can be effected by lobectomy, but on occasions the tumour may be so placed as to necessitate pneumonectomy.

When the tumour is entirely intrabronchial but broad based it is sometimes possible to remove the adenoma along with a portion of the related bronchial wall (D'Abreu and MacHale, 1952). Where the base of the tumour encircles the bronchus a cuff of the bronchus along with the tumour may be excised and

continuity of the bronchus re-established by end-to-end anastomosis (Thomas, 1956). Finally, Bikfalvi and Dubecz (1955) report a case in which an adenoma in the right-stem bronchus involving the right upper lobe was treated by upper lobectomy, plus a cuff of stem bronchus, continuity being re-established between stem bronchus and mobilized lower-lobe bronchus.

If the biopsy tissue is reported as showing a more malignant type of adenoma, particularly a cylindroma, pneumonectomy is the only treatment likely to save the patient (Hazel, Hollinger, and Jensik, 1949).

A good recovery from a successful radical surgical procedure for adenoma usually implies freedom from recurrence or further trouble as the result of the tumour.

In the Massachusetts General Hospital series Harrington, Moersch, Finney, McDonald, and Clagett (1946) reported 88 per cent of adenoma cases alive and well from 3 to 5 years after surgical removal. If the adenoma is of the cylindroma group the prognosis on account of the development of recurrence is much less favourable, being reduced by 50 per cent.

It is as well to remember to bronchoscope the patient at the end of the operation, as not only blood and pus may have escaped into the healthy lung, but also portions of adenoma may have broken off and passed over the carina into the other bronchus.

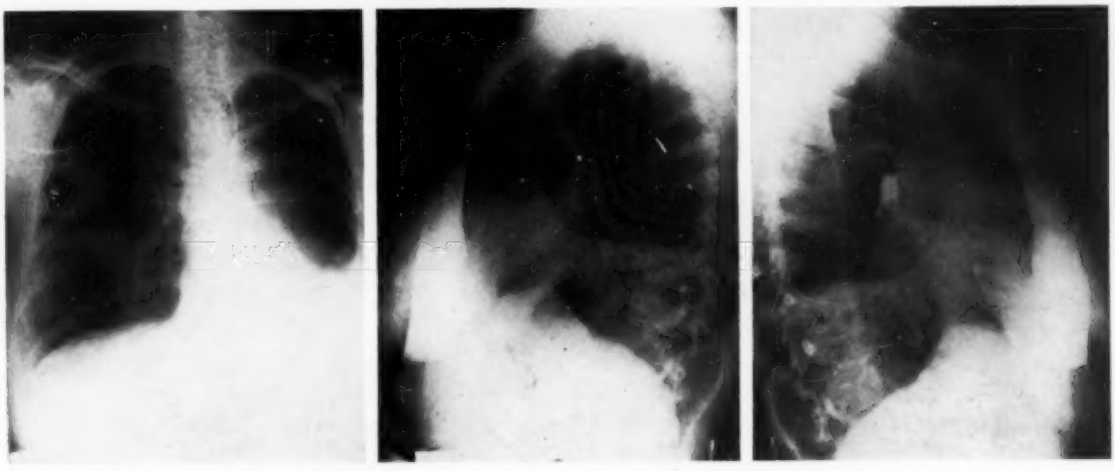
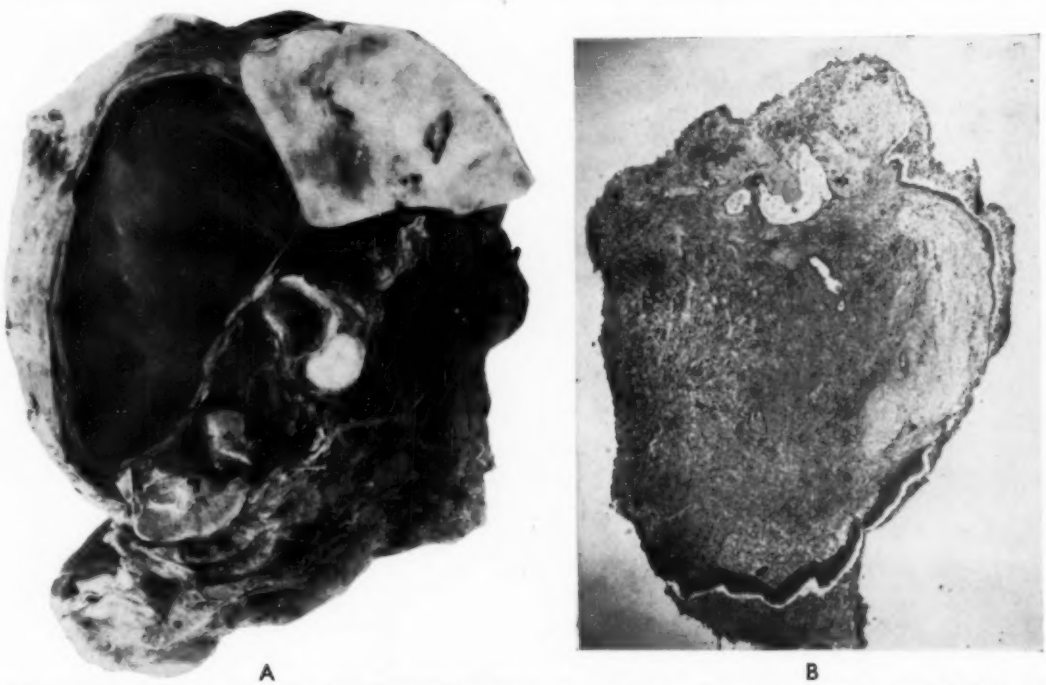


FIG. 702.—*Case* 7. The tumour in this case was 1·5 × 1 cm. and was blocking the left-stem bronchus proximal to the upper-lobe orifice. The tension air cyst measured 12 × 10 cm. Lipiodol outlines the upper edge of the tumour.



A B

FIG. 703.—*Case* 7. A, The tension cyst surmounting the lung apex has been opened. Note the smooth lining. One bronchial opening into the cyst can be seen. The stem bronchus has been opened to show the dark intrabronchial tumour surface. B, A low-power photomicrograph of the tumour.

Clagett and Payne (1946) in 1 case removed from the healthy bronchus a portion of adenoma 2 × 0·5 cm.

CASE REPORTS

Case 1.—For 6 months a 59-year-old woman had had pain in the left chest, lassitude, cough and spit, but no hæmoptysis. She was bronchoscoped in another hospital and the biopsy tissue was reported as oat-cell carcinoma.

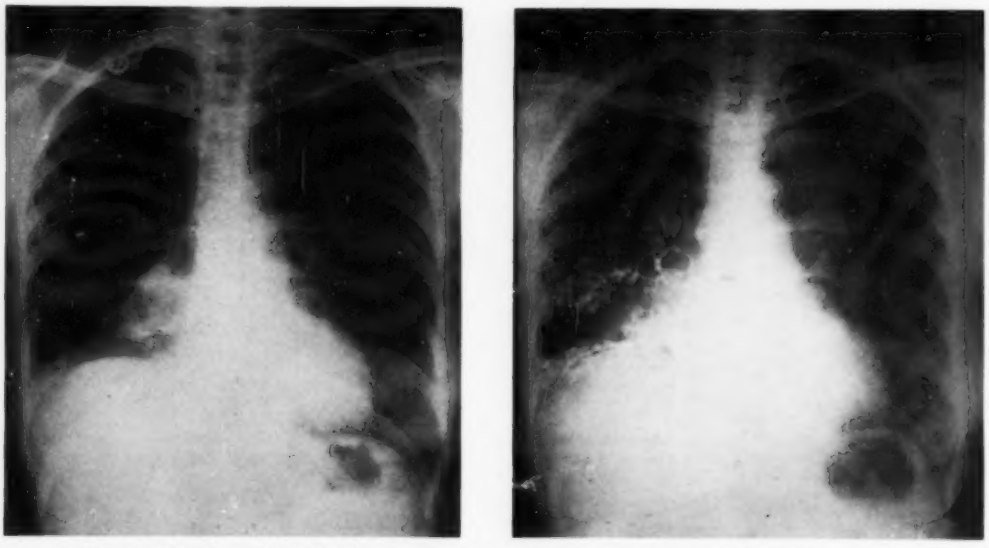
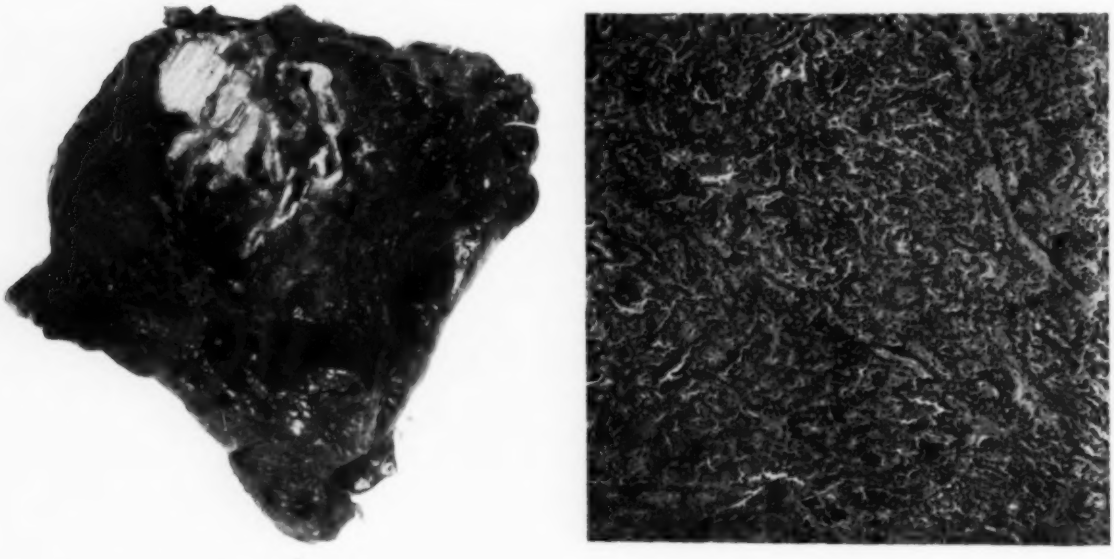


FIG. 704.—*Case* 8. Partial atelectasis of right lower lobe due to an adenoma in the lower-lobe bronchus. There is considerable elevation of diaphragm and incomplete lipiodol filling of the segments of the lower lobe.



A B

FIG. 705.—*Case* 8. A, Section of the lower lobe reveals the rounded white tumour above an area of bronchiectasis. B, The photomicrograph displays the characteristic uniformity of cell arrangement.

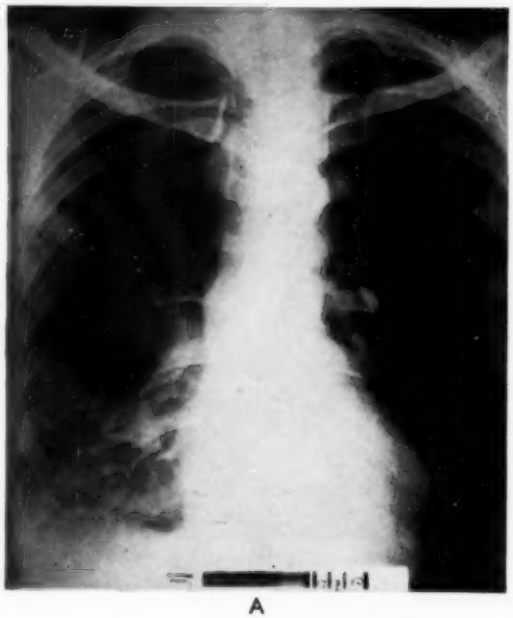
A pneumonectomy was carried out; subsequent examination revealed a polypoidal tumour blocking the stem bronchus. The tumour was an adenoma. (*Fig.* 695.)

Case 2.—A housewife, aged 51, for 2 years had recurring attacks of what appeared to be pneumonia with occasional hæmoptysis. Radiography revealed collapse of the right lower lobe, and bronchoscopy showed an adenoma in the right-stem bronchus. The tumour was removed along with the middle and lower lobes; both lobes were atelectatic, having been blocked by the tumour, which had no extrabronchial extension. (*Fig.* 696.)

Case 3.—A 67-year-old retired shepherd had for 3 years attacks of breathlessness and bronchitis which forced him to give up work. No hæmoptysis. He had crepitations at the left base. Radiographs showed collapse of the left lower lobe. Bronchoscopy showed at the level of the left upper-lobe orifice a ball-valve type of polyp, pink, soft but very tough, and moving with respiration.

AT OPERATION.—The tumour was removed by bronchotomy. The polyp was $\frac{1}{2} \times \frac{1}{2}$ in. and was attached to the posterior bronchial wall by a slender stalk 1 in. in length. Simple adenomatous polyp. It was inferred that the attacks of breathlessness were due to the polyp partly or completely obstructing the stem bronchus. Satisfactory recovery.

Case 4.—Every 6 months for 26 years this 49-year-old housewife had attacks of hæmoptysis. For 3 years she was known to have a simple tumour in the right lower lobe. Shortly before admission she had several large hæmoptyses following one of which she fainted. The tumour was seen and diagnosed on bronchoscopy. It was removed by lower lobe lobectomy. (*Figs.* 697, 698.)



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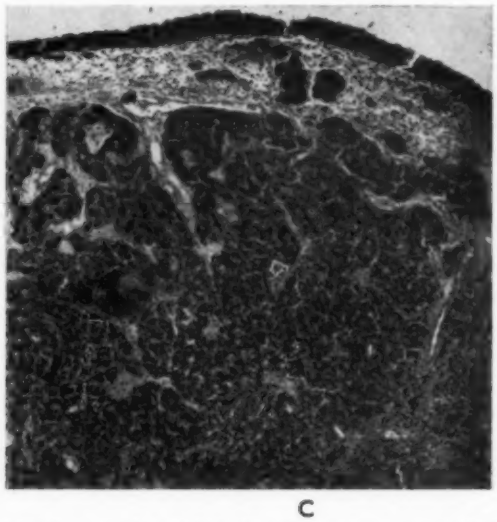

C

FIG. 706.—*Case* 9. A, Adenoma in right lower-lobe bronchus causing atelectasis. The tumour removed through the bronchoscope is shown. B, C, The photomicrographs show the intact bronchial epithelium with a connective-tissue layer intervening between the epithelium and the adenoma.

Case 5.—For 11 years this 46-year-old woman had had a cough with hæmoptysis every 3 or 4 months. For 10 years she was known to have an opacity in the right chest which had altered little in size. An adenoma was seen on bronchoscopy projecting from the middle-lobe bronchus. The tumour was removed along with the middle and lower lobes. (*Figs.* 699, 700.)

Case 6.—Housewife, aged 23. For 1 year she had shortness of breath which was increasing, and she felt wheezy. One hæmoptysis 4 months prior to admission. Pneumonia and left pleurisy 6 years before, and 11 attacks of pleurisy in next 3 years. In sanatorium for 10 months during that time, thought to be a case of tuberculosis. The wheezing was easily heard. Radiographs showed consolidation of left upper lobe. At bronchoscopy the left-stem bronchus was normal as far as the left upper-lobe orifice; at this level tumour tissue was seen projecting into the stem bronchus from the upper-lobe orifice which was blocked by tumour. Biopsy caused much bleeding.

PATHOLOGICAL REPORT.—Bronchial adenoma, like a mixed salivary tumour. The tumour was arising in the bronchial wall and consisted of cells arranged in solid irregular masses or reticulated cords. In parts the cells had formed small acini containing mucin. The tumour cells were uniform and polygonal. Mitotic figures were infrequent. Mucosa showed marked squamous metaplasia.

AT OPERATION.—Pneumonectomy; the upper lobe was solid. The tumour had invaded the bronchial wall and was eroding cartilage. Good recovery. (*Fig.* 701.)

Case 7.—This 30-year-old woman had attacks of pneumonia 5 and 3 years before admission to the thoracic unit. Nine months before admission a pleural effusion was found. Two months before admission the effusion was found to be due to sterile pus. It had caused complete lung atelectasis and it was lying in a space which had a slightly positive pressure. Several attempts were made to aspirate air and re-expand the lung, but these were unsuccessful. Bronchoscopic examination revealed an adenoma. Two more bronchoscopic examinations were carried out and further tumour tissue was removed but lung re-expansion did not occur. Pneumonectomy was performed and a large tension lung cyst of the upper lobe was found. The fluid and subsequent sterile pus had apparently been aspirated from the cyst and not from the pleura. The specimen showed an adenoma 1 × 1·5 cm. blocking the stem bronchus above the origin of the upper-lobe bronchus. The cyst was 12 × 10 cm.; it had at least three communications with the upper-lobe bronchi. (*Figs.* 702, 703.)

Case 8.—This 49-year-old woman had recurring hæmoptysis for 10 months. Bronchoscopy revealed an adenoma in the right lower-lobe bronchus. Radiographs of the chest following one brisk hæmoptysis revealed an increase in opacity in the lobe probably due to aspiration of blood. The tumour and right lower lobe were removed. (*Figs.* 704, 705.)

Case 9.—A motor driver, aged 39, 2 months before admission to the thoracic unit, developed what was thought to be influenza, following which he had a hæmoptysis of 1 pint of blood. He was bronchoscoped in another hospital and the biopsy specimen was reported as oat-cell carcinoma. Bronchoscopy was repeated when he was admitted to the thoracic unit and an adenoma was seen and removed. Subsequently lobectomy was carried out, and complete removal of the tumour at bronchoscopy was confirmed. (*Fig.* 706.)

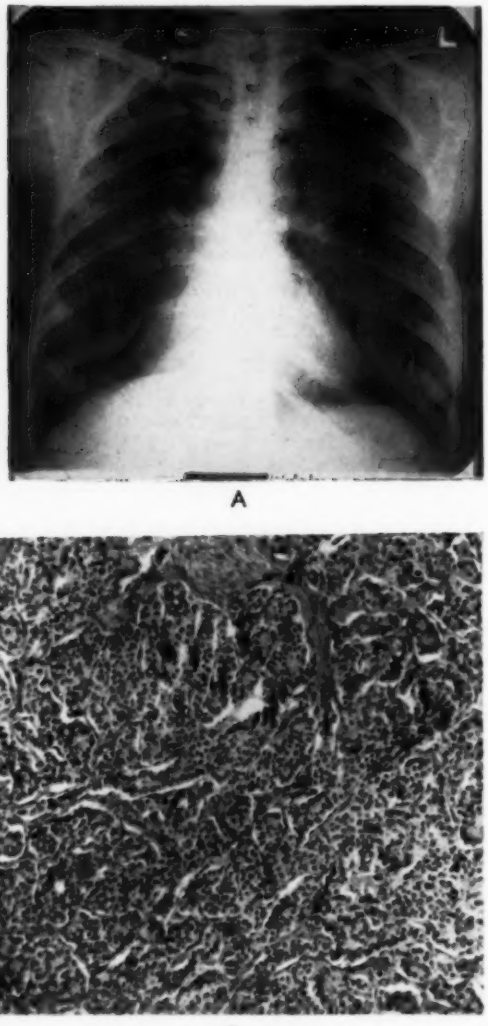

FIG. 707.—*Case* 10. A, B, Partial collapse of right middle and lower lobes due to the intrabronchial adenoma shown as it lies just below the upper-lobe orifice (the stem bronchus has been laid open). Bronchial biopsy in this case was reported as oat-cell carcinoma (C), hence the reason for pneumonectomy.

Case 10.—A 42-year-old domestic servant had been breathless on exertion for 20 years; for 6 years a ticklish cough had been present. Two attacks of so-called pneumonia occurred, the first 2 years and the second 1 month before admission to the thoracic unit. Bronchography revealed incomplete obstruction of the right-stem bronchus just above the middle-lobe bronchus. Bronchoscopic biopsy tissue taken in Aberdeen before admission was reported as oat-cell carcinoma. Pneumonectomy was carried out and the diagnosis was confirmed by an inexperienced pathologist. Three years later she was admitted to Perth Royal Infirmary for some other condition. She was so well that a report was sent to the thoracic unit telling of her good health. The specimen and histological sections were reviewed and it was found that the tumour was an adenoma and not a carcinoma. (*Fig.* 707.)

Case 11.—For 10 years a 29-year-old shop assistant had had frequent bouts of influenza. Seven years prior to admission she had had pleurisy and pneumonia. Subsequent to this illness she had frequent attacks of coughing with varying amounts of sputum: no hæmoptysis. Three months before admission she had an attack of influenza, and following this illness she felt very tired. She had increasing breathlessness for 1 year. The left side of the chest was flattened and moved poorly on respiration. She was dull over the left side. Respiratory murmur and vocal fremitus were diminished on the left side, especially over the left lower zone. Radiographs showed a grossly shrunken lung which was opaque; the heart was markedly displaced to the left. A large translucency in the left clavicular region posteriorly

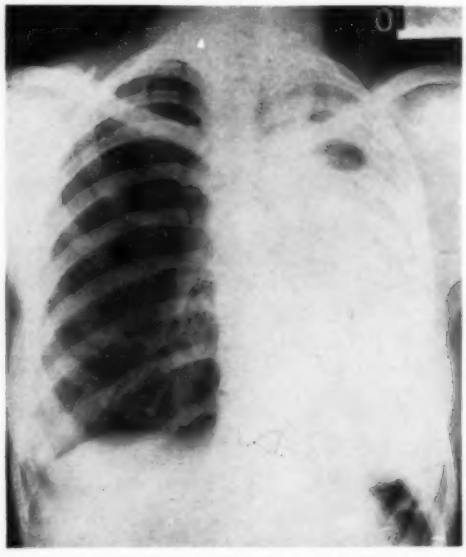

FIG. 708.—*Case* 11. Complete atelectasis of left lung due to obstruction from an adenoma of upper lobe blocking the stem bronchus. The trachea and mediastinum are deviated to the left. There is a cavity in the upper lobe.

suggested a large cavity containing a little fluid. Bronchoscopic examination revealed the carina to be twisted clockwise. A large, red, smooth polypoid tumour projected from the left upper-lobe bronchus. Biopsy revealed an active bronchial adenoma, medium sized, rounded, compact masses of small cuboidal cells. Pneumonectomy. Satisfactory recovery. (*Fig.* 708.)

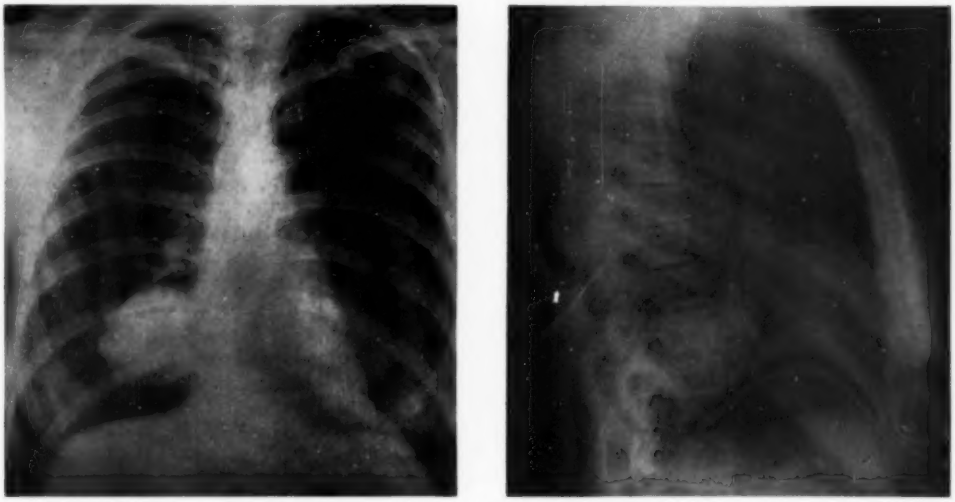

FIG. 709.—*Case* 12. The rounded shadow in the right lower lobe was removed by lower-lobe lobectomy. Initially the tumour was believed to be an adenocarcinoma, but at a later review it was confirmed as an adenoma.

Case 12.—Two months prior to admission this female, aged 38, developed a chest cold during which she had several hæmoptyses. Examination revealed dullness at the right lung base. Radiography showed a rounded solid shadow at this area. She was operated on and the tumour, along with the right lower lobe, was removed. It was found to be 5 cm. in diameter. There was some doubt as to the exact diagnosis, but it was thought to be an adenocarcinoma. She made a satisfactory recovery but died 5 years later from a carcinoma of the breast.

On reviewing the case and taking into account her subsequent good health, it would seem that it was an adenoma and not an adenocarcinoma. (*Fig.* 709.)

CONCLUSION

At one time adenoma of the bronchus was looked upon as an extreme rarity; indeed, Dick (1950) was unable to find an example of this type of tumour in the post-mortem records of three large Glasgow teaching hospitals, and yet here in one surgical thoracic unit are 12 proven cases occurring in the space of 13 years. Dick reported 11 personal cases, including 9 of the cases here described, the other 3 cases reported in this paper being operated on by Dick subsequent to 1950. It seems that advances in radiography and the advent of bronchoscopy, and latterly, surgical exploration, will bring an ever-increasing number of adenoma tumours to light.

In former years it is probable that these cases either died at home under the mistaken diagnosis of unresolved pneumonia or carcinoma, or that they died in sanatoria, erroneously diagnosed as cases of tuberculosis.

SUMMARY

An account is given of the clinical, radiological, and pathological findings in 12 cases of carcinoid type of bronchial adenoma. The treatment of adenoma is discussed.

Acknowledgement.—I wish to record sincere thanks to Mr. Bruce M. Dick for permission to publish these cases and for his encouragement in the preparation of this paper. My thanks are also due to Mr. Robert McLean for the photographs.

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SPONTANEOUS SUBCORTICAL HÆMORRHAGE IN YOUNG ADULTS

A REPORT OF 7 SURGICALLY TREATED CASES

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INTRACEREBRAL hæmorrhage is generally considered to be very uncommon before the age of 35 years. Certainly after this age the liability increases with each decade. The condition is most common between 50 and 65 years and is then a frequent terminal event. Cerebral hæmorrhage, however, is occasionally encountered in early adult life (Duncan, 1936; Kidd and Cummings, 1947), and even in infancy and childhood (Sheldon, 1927). This report is based upon 7 cases of intracerebral hæmorrhage, in patients between the ages of 19 and 34 years, treated surgically between April, 1953 and December, 1955. Klemme (1941) suggested that the apparent low incidence of non-traumatic subcortical hæmorrhage with clot formation might be due to lack of recognition of the syndrome, or to the infrequency of the condition. The occurrence of these cases of verified spontaneous intracerebral hæmorrhage in young adults, in a relatively short space of time, would suggest that the condition is perhaps not so uncommon as is generally believed.

AETIOLOGY

The aetiology of spontaneous intracerebral hæmorrhage has been discussed in some detail by Jewesbury (1947), but the exact underlying cause, in many cases, remains in doubt. In the elderly, hypertension and arteriosclerosis account for the majority of cases, while the intracerebral rupture of basal aneurysms can be held responsible for a few. The latter condition is a more common cause of cerebral hæmorrhage in young adults. The cases reported here, however, were not associated with basal aneurysms and indeed, with one exception, angiography failed to show any vascular malformation. Hypertension was absent, and although arterial degeneration may occur at an early age in patients with this predisposition, there was no evidence of arterial disease in any of these patients on clinical examination. Syphilis was excluded in all but one case by serological tests. There was no history of head trauma, and contrary to the cases described by Craig and Adson (1936) no history of violent exertion or severe emotional strain, although *Case* 1 must undoubtedly have been associated with a certain degree of excitement.

In *Case* 5 angiography revealed early filling of a solitary vein in the left parietal region (*Fig.* 711). This raised the question of a small congenital angiomatous malformation and, in this particular case, this seems the most likely explanation of the cerebral hæmorrhage, although there was no previous history to suggest such a lesion. Hæmorrhage, however, is a recognized complication of these congenital malformations and may well be the presenting symptom. It is also not uncommon for small angiomas to show no greatly enlarged arteries, but only distended veins seen in the arterial or capillary phase on angiography. The lesions also appear to occur most frequently in the distribution of the middle cerebral artery (Bailey, 1948).

The fact that the vascular abnormality in *Case* 5 could not be demonstrated on repeat angiography, after the recovery of the patient, may be of some significance in the question of aetiology in these cases in general. It is possible that following the rupture of the angioma, with the formation of the intracerebral clot, partial destruction of the lesion occurred and this was followed by thrombosis in the remainder. Operative experience with these malformations (Norlén, 1949) has shown that if the arteriovenous shunt is removed then the secondarily dilated vessels will revert to normal. Disruption of the shunt as the result of hæmorrhage would presumably have the same effect. Potter (1955) described a case in which autopsy failed to reveal an angiomatous malformation of some size, in a patient who died from cerebral hæmorrhage 6 years after the lesion had been demonstrated at operation. It would seem even more likely that a small malformation could be completely destroyed at the time of rupture and so fail to be demonstrated by angiography or even at autopsy.

Cases of spontaneous intracerebral hæmorrhage, in which the cause has been shown to be due to the rupture of a small arteriovenous malformation, have been reported (Kidd and Cummings, 1947; Russell, 1954). Margolis, Odom, Woodhall, and Bloor (1951) reported 4 cases of fatal cerebral hæmorrhage bearing a close resemblance to spontaneous intracerebral hæmorrhage of unknown aetiology. In 2 of these cases the causative lesion, a vascular malformation, was demonstrated only on histological examination of the wall of the hæmatoma, while in the other 2 cases the angiomatous lesion responsible for the hæmorrhage was found to be small and deeply situated. In the same paper 2 cases with small vascular lesions, found incidentally, and which had not given rise to gross hæmorrhage, were reported. It was thought that these lesions were so small that, had massive hæmorrhage occurred, great difficulty would have been experienced in locating the cause. The authors expressed the view that small angiomatous malformations might play a more significant role in spontaneous cerebral hæmorrhage than was usually recognized. It is interesting to note that Potter (1955) in a recent review of cases of angiomatous malformation pointed out that it was the small lesion which showed the greatest tendency to rupture and bleed.

The arterial theory of intracerebral hæmorrhage is commonly accepted (Alpers, 1946), but there is evidence to suggest that, in some cases, the rupture in the vascular system appears to occur on the venous side. This has been pointed out by Craig and Adson (1936), and is possibly a further factor which suggests

a small angioma as a cause of this syndrome. Werner (1954), nevertheless, expressed the view that slow venous oozing could hardly be responsible for the apoplectic onset observed. A striking feature, however, of the cases forming the basis of this report was the fact that the dramatic onset usually associated with adult apoplexy did not occur. The initial symptoms were of sudden onset, but there was no loss of consciousness, unless epilepsy occurred, and there was a gradual progression of symptoms over a period of hours and in some cases several days. The sudden, but not apoplectic, onset can be explained by the fact that hæmorrhage occurring from the venous side of an angiomatous malformation would certainly be more brisk than ordinary venous bleeding, but would lack some of the destructive power of arterial hæmorrhage. In these cases the extravasation of blood into the hemisphere was not very extensive, as would be expected in arterial bleeding.

The following cases are reported to stress again the existence of this condition which, as pointed out by Werner (1954), appears to exist as a clinical entity. They illustrate the relatively good prognosis, when treated surgically, both as regards the survival of the patient and the final residual disability.

CASE REPORTS

Case 1.—Miss V. J., aged 22 years, was preparing for her wedding on the following day when on the evening of April 3, 1953, she was taken suddenly ill. She lost the power of speech and was unable to use the right arm or leg. At about the same time she developed a severe generalized headache. A few hours later she became drowsy and vomited; there was no loss of consciousness and no epileptic convulsions. The following day she was admitted to a local hospital in a somewhat improved state, headache persisted, and she complained of a pins and needles sensation in the right arm and leg.

There was nothing relevant in her personal or family history.

The patient was transferred to the neurosurgical unit at the Cardiff Royal Infirmary on April 9, 1953.

On Examination.—She was conscious but rather drowsy. There was an expressive dysphasia present with a right lower facial weakness and a complete right flaccid hemiplegia. The tendon reflexes on the right were absent, the plantar response was extensor. Cortical sensation was impaired on the right side of the body. The fundi were normal. B.P. was 140/85, blood W.R. was negative. The cerebrospinal fluid was under a pressure of 250 mm., the protein 31 mg. per cent, and the fluid was slightly xanthochromic.

A left carotid angiogram on April 11 showed a minimal shift of vessels indicating a space-taking lesion in the left frontoparietal region. No aneurysm or other vascular abnormality was detected.

An E.E.G. showed some generalized fast activity; no focus could be demonstrated.

At Operation.—On April 15 a left parietal burr-hole was made and a substantial amount of dark fluid blood aspirated from deep in the hemisphere.

Post-operative Progress.—Some recovery of function was apparent in the right arm and leg by April 27, and on discharge from hospital on May 13 there was only a slight right hemiparesis present, but with marked astereognosis in the right hand. Improvement continued and when last seen on Dec. 8, 1955, the patient was very well and without complaints. There was a very minimal right hemiparesis present with slight cortical sensory impairment in the corresponding hand. The tendon reflexes were increased on the right. Fundi were normal and the visual fields were full. She was leading a normal life in every respect. On leaving hospital the unavoidably delayed wedding took place, and on this last follow-up examination she was 7 months pregnant.

Case 2.—Mrs. E. B., aged 33 years, was admitted to the Royal Infirmary, Cardiff, as an emergency on June 12, 1954. About 45 minutes before admission the patient, who had previously been quite well, complained of numbness and inability to move the left arm. She felt sick, vomited, and was put to bed. There followed a tremor of her limbs, her face became twisted, and she lost consciousness. There was no complaint of headache. There was nothing of significance in the past medical history. She was 3 months pregnant.

Fig. 710.—*Case* 2. Right cerebral angiogram showing slight stretching of vessels in the parietal area.

On Examination.—She was unconscious, the right pupil was larger than the left and there was a complete left hemiplegia. The fundi were normal. There was no neck rigidity at first, but this became marked after 24 hours, and lumbar puncture showed a blood-stained fluid under a pressure of 290 mm. She was transferred to the neurosurgical unit on June 23. At that time she was conscious but drowsy. The left hemiplegia remained complete and there was in addition a marked left hemianæsthesia. B.P. was 130/80, blood W.R. and Kahn were negative.

A right carotid angiogram on June 23 showed no aneurysm or other vascular abnormality, but there was a shift to the left of the anterior cerebral artery, and slight stretching of vessels in the parietal region (*Fig.* 710).

At Operation.—On June 30 a right frontal craniotomy was performed. An intracerebral hæmatoma was found in the frontoparietal region. When the clot had been cleared out it was possible to see that the cavity communicated with the lateral ventricle. The cavity, which measured 6 × 6 × 4 cm., did not extend medially to the midline, nor did it extend downwards as far as the sylvian fissure. No vascular malformation could be found in the wall of the hæmatoma.

Post-operative Progress.—This was uneventful. At the time of discharge from hospital on July 22 the patient could walk with help, there was a little voluntary power at the left elbow, but otherwise she could not use the left arm or hand. In November her child was delivered by Cæsarean section. Just prior to this she had a major epileptic convulsion and a further fit occurred in January, 1955. When last seen on Jan. 27, 1955, there had been further improvement. She was able to walk with the aid of a leg calliper and there had been some more return of function to the left upper limb. There remained, however, a severe hemiparesis with considerable

sensory impairment; in spite of this she was able to carry out her household duties well.

Case 3.—Mrs. S. L., aged 23 years, was perfectly well until the morning of March 5, 1955. She suddenly became aware of a feeling of pins and needles in the left side of her face and a peculiar sensation in the head which she could not describe. Her speech became slurred. Within an hour the pins and needles sensation spread to involve the whole of the left side of the body. She collapsed but did not lose consciousness. The left arm and leg were weak and lacked normal sensation. She developed a mild frontal headache and vomited. She was admitted to a local hospital and subsequently transferred to the neurosurgical unit at Cardiff on Jan. 12, 1955. There was nothing relevant in her personal or family history.

On Examination.—She was drowsy and neck rigidity was present. There was a severe degree of left facial weakness present and a flaccid left hemiparesis which was complete in the arm and almost so in the leg. In addition there was complete left hemi-anæsthesia. The lumbar cerebrospinal fluid was under normal pressure and there was no abnormality in the cytology or chemistry. The C.S.F. and blood W.R. were negative; B.P. was 120/90.

A right carotid angiogram on March 19 showed no aneurysm or other vascular abnormality, but revealed a space-occupying lesion in the right frontoparietal region.

At Operation.—On March 19 a right lateral craniotomy was performed. Deep to the motor cortex an intracerebral collection of blood was located on needling. Some 30 c.c. of dark altered blood were aspirated. In view of the site of the lesion it was decided not to make a cortical incision.

Post-operative Progress.—This was uneventful. Some evidence of recovery of function was present by March 24. When last seen on Dec. 22, 1955, the patient was well and free from complaints except for occasional right frontal headache. Power in the left leg was good, and she was able to walk on her own without assistance but with a spastic gait. There had been some recovery of function in the left arm and gross movements were possible, including good finger flexion. Cortical sensory loss was still marked in both the arm and leg. In spite of her disability she was capable of doing her own housework, and looked after her two children.

Case 4.—Mr. V. S., aged 28 years, was perfectly well up to June 7, 1955. On that morning he returned home from work and went straight to bed, as though in a trance, without speaking to his wife. People with whom he worked said that he had suddenly appeared ill, and had complained of some slight headache before he left work. His personal and family history were not relevant. He was admitted to hospital in the early hours on June 8.

On Examination.—He was conscious, drowsy, and somewhat unresponsive. There was an expressive dysphasia present. Neck rigidity was present and Kernig's sign positive. He moved all his limbs equally, but the tendon reflexes were more brisk on the left than the right. The plantar responses were flexor and there was no apparent sensory abnormality. His fundi were normal. B.P. was 120/70. The lumbar cerebrospinal fluid was under a pressure of 300 mm., was slightly blood stained, protein 158 mg. per cent, and the W.R. was negative.

The E.E.G. showed frequent intermittent runs of high voltage theta and delta waves in all leads, but a more persistent delta component occurred focally in the left frontal region.

A left carotid angiogram showed no evidence of an aneurysm or other vascular abnormality, but there was a shift of the anterior cerebral artery to the right of the midline.

At Operation.—On June 13, 1955, a left frontal craniotomy was performed. An intracerebral hæmatoma was found in the posterior part of the frontal lobe. The clot, which was mainly solid, was removed. The resulting cavity measured 7 × 4 × 5 cm. and may have communicated with the anterior horn of the lateral ventricle. The cortex on the medial side of the cavity was intact. The middle cerebral artery was explored from the carotid bifurcation to the insula. No vascular abnormality was demonstrated.

Post-operative Progress.—This was uneventful and the patient went on to make a complete recovery. When last seen on Sept. 29, 1955, he was without complaint, and had returned to his original occupation of engineering. There was no dysphasia, and with the exception of a slight increase in the tendon reflexes in the right arm and leg, no abnormal neurological signs were demonstrated.

Case 5.—Mrs. M. F., aged 19 years, was quite well until Dec. 22, 1955. On that day while out shopping she suddenly experienced a peculiar sensation in the frontal region of her head. She dropped her shopping basket, but did not lose consciousness. The movements of her right arm became clumsy and she was unable to speak clearly. She reached home and went straight to bed. The following day she had pain in the occipital region and back of the neck. She could say a few words, but was unable to give a clear account of her illness. She was admitted to a local hospital on Dec. 24. There was some improvement in her speech during the following few days, but she soon began to show evidence of raised intracranial pressure with headache, vomiting, and the development of papillœdema. There was nothing relevant in her past history. Her mother had developed a hemiplegia 12 months previously which was thought to have been due to a cerebral embolus. She was transferred to the neurosurgical unit of the Cardiff Royal Infirmary on Dec. 28, 1955.

On Examination.—The patient was fully conscious and co-operative, but very distressed by her dysphasia. She had great difficulty in expressing herself, but was able to understand simple commands. There was marked apraxia. Very gross bilateral papillœdema was present with hæmorrhages, and she was experiencing amblyopic attacks. On confrontation there was a right homonymous hemianopia affecting the upper quadrants more than the lower. There was a left sixth nerve paresis, a slight lower right facial weakness, and a little weakness of the right arm and leg. Sensory testing revealed marked impairment to all modalities over the right side of the body. B.P. was 110/60, blood W.R. and Kahn were negative.

An E.E.G. showed some high voltage irregular delta activity focally in the left temporal region.

A left carotid angiogram showed a slight shift of vessels indicating a space-occupying lesion in the left parietal area. In addition there was a small focal vascular abnormality in the left parietal region (*Fig.* 711).

On Jan. 1, 1956, the patient had a major epileptic attack; following this episode she was rather drowsy, vomited, and complained of increasing headache.

At Operation.—On Jan. 4 a left parietal burr-hole was made. The brain appeared normal externally as far as could be seen. Thick fluid blood was encountered at a depth of 5 cm. A small cortical incision was made and a considerable amount of solid clot removed by suction. A post-operative radiograph was taken to show the site and extent of the intracerebral cavity (*Fig.* 712).

Post-operative Progress.—The patient's immediate post-operative condition was quite satisfactory, she was more alert, and free from headache. Three days later, however, the left sixth nerve paresis became complete, but following this, recovery progressed uneventfully. She was discharged from the unit early in February. At that time her speech had improved markedly. She was

able to carry on a simple conversation with ease and there was no obvious apraxia. Testing, however, still showed some degree of both expressive and receptive dysphasia. Ocular movements were full and there was no subjective diplopia. The visual fields were almost full on confrontation and the papillœdema was much less marked. No muscular weakness could be detected in the extremities, but there was still cortical sensory loss over the right side of the body, although less marked than on admission.

Left carotid angiography was repeated before her discharge from hospital. The small focal vascular abnormality seen in the original pictures was not demonstrated in the views taken.

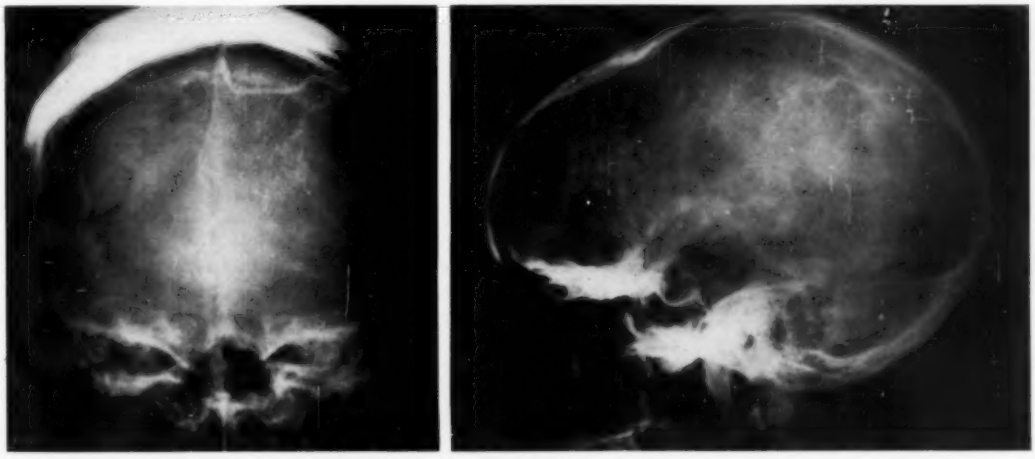

FIG. 711.—*Case* 5. Anteroposterior and lateral left cerebral angiogram showing small angiomatous malformation in the parietal area.

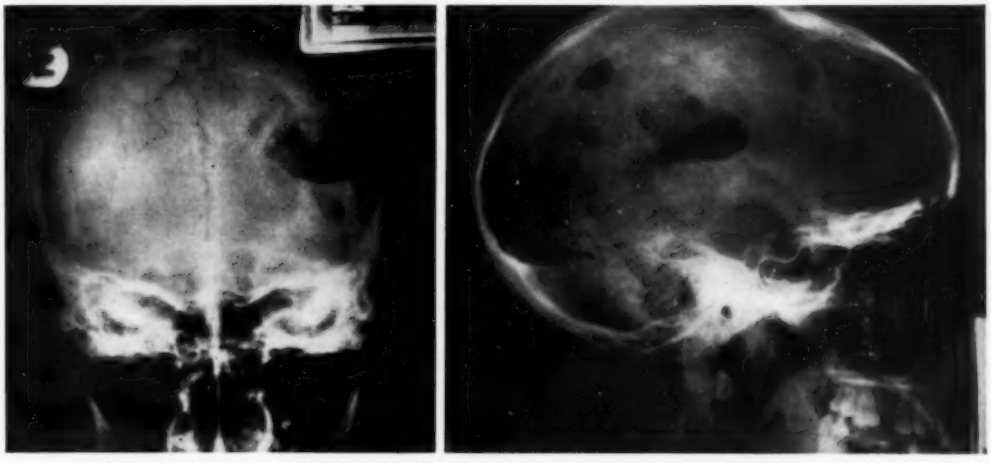

FIG. 712.—*Case* 5. Anteroposterior and lateral views showing air-filled hæmatoma cavity.

Case 6.—Mr. I. B., aged 34 years, was perfectly well until Aug. 3, 1954. On that day, soon after rising from bed in the morning, he suddenly lost consciousness in a major epileptic attack. This was the first of several such attacks. He was admitted to a local hospital where neurological examination failed to reveal any abnormal signs. On Aug. 10 he complained of mild occipital headache and vomited. Bilateral early papillœdema was noted and also weakness of the right arm and leg. There was nothing relevant in the past medical history, but in the family there was a history of 'cerebral thrombosis' in the patient's father and uncle. On Aug. 14 he was transferred to the neurosurgical unit at the Morriston Hospital.

ON EXAMINATION.—He was fully conscious, but retarded and concentration was poor. There was a slight degree of expressive dysphasia present, early bilateral papillœdema, a slight right lower facial weakness, and a moderate right hemiparesis most marked in the leg. There was no impairment of sensation. B.P. was 130/90, and blood W.R. was negative.

A left carotid angiogram was performed on Aug. 16. This showed a slight shift of vessels indicating a space-occupying lesion in the left parietal region. No aneurysm or other vascular abnormality was demonstrated.

AT OPERATION.—On Aug. 23 a left frontoparietal craniotomy was performed. The brain was under increased tension and the gyri in the region of the left motor cortex were widened. Needle exploration revealed a collection of dark altered blood deep to this region. A quantity was aspirated but in view of the situation of the lesion the cortex was not incised.

POST-OPERATIVE PROGRESS.—This was uneventful; some recovery of function was present before discharge from the unit on Oct. 10. When last seen on April 10, 1956, he was very well. Recovery of function in the right arm and leg was almost complete; there was just a little weakness in the lower limb and slight impairment of position sense in the toes. The tendon reflexes in the right arm and leg were more brisk than those on the left. Since his discharge from hospital he has had occasional epileptic attacks, but these were well controlled with anticonvulsants. He was working full time.

Case 7.—Miss M. M., aged 20 years, was perfectly well until Dec. 16, 1954. On that day while washing her face at work she suddenly developed a mild occipital headache. The headache increased in severity over a matter of 10–15 minutes, until it became very severe and she vomited. There was no clouding of consciousness. She was taken home and put to bed. There was some improvement in the headache over the next 3 days, but this was followed by a further attack of severe headache which tended to be right sided and lasted for over a week. She then improved a little, but on Dec. 28 headache and vomiting were again severe and she had an attack which her doctor thought was epileptic in nature. On recovering consciousness she complained of severe left-sided headache and of weakness in the right arm and leg. On Dec. 30 she was found to have papilloedema, and a lumbar puncture, performed at a local hospital, showed that the cerebrospinal fluid was under a pressure of 240 mm. and was xanthochromic. There was nothing relevant in her past medical history. Her father had had several 'blackouts' following a fall from a ladder.

On Jan. 2, 1955, she was admitted to the Morriston Neurosurgical Unit.

On Examination.—She was drowsy and uncooperative. There was no obvious dysphasia, bilateral papilloedema was present and this increased rapidly in intensity in the following few days. The visual fields were full on confrontation, conjugate deviation of the eyes to the left was impaired. There was a marked right hemiparesis present affecting the lower limb more severely than the upper limb. Tendon reflexes on this side were increased and the plantar response was extensor. There was no impairment of sensation. B.P. was 120/70.

The E.E.G. was grossly abnormal with outbursts of slow activity in both hemispheres, but with some phase reversal indicating a left-sided lesion.

At Operation.—On Jan. 7, 1955, a left frontoparietal craniotomy was performed. The brain was under increased tension and the convolutions in the left motor cortex were widened. On needling deep to the motor area an intracerebral collection of dark altered blood was encountered. A considerable quantity was removed by aspiration and this resulted in lessening of the brain tension.

Post-operative Progress.—This was uneventful. By Jan. 14 there was some recovery of function in the right arm and leg. When last seen on April 17, 1956, she was well and free from complaints. She had returned to work in July, and had married in September, 1955. Recovery of function was almost complete. No weakness could be detected in the upper limb and the tone was normal; the lower limb was slightly weak with increase of tone. The tendon reflexes on the right were exaggerated and the right plantar response was still extensor. There was no abnormality of sensation. The visual fields were full on confrontation. She was leading a completely normal life, her only disability being difficulty in running owing to the stiffness of the right leg. There had been no further blackouts.

This was the only case in the series in which angiography was not done. The clinical picture strongly suggested a spontaneous intracerebral hæmorrhage, and in view of the rapid progression of the papilloedema and the deterioration in her general condition, exploratory craniotomy was undertaken without contrast studies. The operative findings confirmed the clinical diagnosis and, therefore, inclusion of the case in this series seems justified.

DISCUSSION

Incidence.—It is not possible, in view of the small number and the highly selective nature of these cases, to come to any conclusions regarding the age or sex incidence of this condition. It seems that typical cases of spontaneous intracerebral hæmorrhage with clot formation are more common in relatively young patients. This is supported by published cases (Penfield, 1933; Craig and Adson, 1936; Klemme, 1941; Pilcher, 1941; Effron, Rosenberg, and Muller, 1949; and Grant and Austin, 1950). Only cases under the age of 35 years have been considered in this paper in an attempt to define more clearly the clinical entity of spontaneous subcortical hæmorrhage. In the older age groups, in which there can be no doubt that the condition does occur, vascular degenerative disease and hypertension play an increasing role in the cause of cerebral hæmorrhage, and so tend to complicate the clinical picture. Werner (1954) stated that the condition was three times more common in men than in women. In this series there were 5 women and only 2 men.

Location.—Unlike ordinary cerebral hæmorrhage, which usually occurs in the region of the internal capsule and basal ganglia (Boyd, 1943), the site of the spontaneous intracerebral hæmorrhage under discussion has usually been found to lie relatively near the surface (*Fig.* 712). Hæmorrhage in this series of patients occurred into the left hemisphere on 5 occasions and into the right hemisphere on 2. Klemme (1941) collected 17 surgically treated cases from the literature, and added 6 of his own; in this series the left hemisphere was also more commonly involved as it was in the cases reported by Pilcher (1941). It is not known whether this apparent predominant involvement of the left hemisphere is significant. The posterior temporal or parietal region of the brain is the usual site of the hæmorrhage. In the cases reported here 6 of the clots were found in the parietal and 1 in the frontal region. Spontaneous hæmorrhage occasionally occurs into the cerebellum (Bagley, 1932; Pilcher, 1941).

Symptomatology.—The most important feature of the clinical picture in these cases was the sudden onset of the illness and the gradual progression of the symptoms. There were no prodromal symptoms or predisposing factors. The patients, apparently in perfect health, were suddenly struck down. In spite of this sudden onset, however, the dramatic picture usually associated with adult apoplexy did not occur, and there was no loss of consciousness unless epilepsy supervened. In all but one patient (*Case* 2) headache was present in some degree in the early stages of the illness, but in only 2 patients (*Cases* 1 and 7) was this headache severe and incapacitating. Vomiting occurred as an early symptom in 4 of the 7 patients, and in 2 others vomiting developed during the course of the illness; only 1 patient (*Case* 4) remained free from this symptom. Focal disturbance of cerebral function was an early and often presenting feature of the condition. In all but one (*Case* 4) hemiparesis, of varying severity, was a prominent sign and was frequently associated with a disturbance of sensation. In this series dysphasia was also commonly present. Raised intracranial pressure occurred in all but one patient (*Case* 3) as shown by the presence of papilloedema or raised pressure on lumbar puncture. In 2 cases (*Cases* 5 and 7) papilloedema was both gross and progressive, and it is certain that in the absence of surgical intervention both of these patients would have died from raised intracranial pressure. Definite

clinical evidence of meningeal irritation was detected in only 3 patients. Epilepsy, as an early feature, occurred in 2 patients (*Cases* 2 and 6). In the first the onset was probably focal in the face and arm and then became generalized; in the second major attacks occurred without evidence of focal onset. In 2 cases (*Cases* 5 and 7) there was one isolated attack of epilepsy during the course of the illness. Several of the patients, however, complained of paræsthesiæ in the affected limbs.

Diagnostic Procedures.—In 4 of the 5 patients who had a lumbar puncture performed there was evidence of some bleeding into the cerebrospinal fluid; in one case the fluid was quite normal. In this series straight radiographic examination of the skull did not help in the diagnosis, and air studies were not carried out. Angiography was performed in all but one patient. This investigation would appear to be the one of choice and it seems to carry little risk to these patients. The location of the intracerebral collection of blood was usually revealed, although the vascular displacement was in most cases only slight, and the presence or absence of a vascular abnormality was demonstrated. In this series a vascular abnormality was demonstrated in only 1 patient (*Case* 5; *Fig.* 711); a reason for this has been suggested. It is of course possible that more of these lesions were not shown on angiography because of their microscopic size. The use of serial angiography (Curtis, 1949), however, might prove useful in demonstrating a higher proportion of these small angiomas. In those cases in which electroencephalography was carried out the record was found to be grossly abnormal, but accurate localization of the lesion was not always possible.

Operative Procedures.—Penfield (1933) suggested that in a small number of cases there was a place for surgery in the treatment of intracerebral hæmorrhage and that, with careful selection, this number might be increased. Since then there have been a number of reports of successfully treated cases (Craig and Adson, 1936; Ecker and Love, 1939; Pilcher, 1941; Klemme, 1941; Grant and Austin, 1950; and Werner, 1954); but nevertheless the success of surgical intervention in some cases of cerebral hæmorrhage is not generally recognized. Boldrey (1951) in a discussion on apoplexy in young adults does not mention the possibility of surgical evacuation of the hæmatoma.

The extent of the recovery of function following operation suggests that, while there must be some destruction of brain parenchyma by the hæmorrhage, some of the paralysis is due to the local irritant and pressure effects of the clot following separation of cerebral tissue. The removal of the clot should, therefore, hasten recovery and make this more complete. Some of the patients, after an interval, show a progressive rise of intracranial pressure, as in the case of 2 patients in this series. The mechanism of this secondary rise in pressure has been discussed by Rowbotham and Ogilvie (1945). Here surgical intervention is urgently called for.

The choice of operation rests between simple aspiration of the fluid part of the hæmatoma through a burr-hole, or craniotomy and the complete evacuation of the clot under direct vision. The latter is the operation of choice and gives the patient the best chance of recovery. However, many of these hæmorrhages occur into the dominant hemisphere and the situation is frequently such that a cortical incision would increase the neurological disability. That aspiration of the fluid part of the hæmatoma can be very successful is illustrated by several of the cases reported here. Aspiration may have to be repeated as suggested by Bagley (1932). In none of these cases did bleeding recur into the hæmatoma cavity after aspiration, and in those cases treated by evacuation of the clot under direct vision hæmostasis did not present a problem, as no active arterial bleeding was encountered. Decompression should not be necessary if all or even some of the hæmatoma is removed.

Prognosis.—The life expectancy of patients with this condition is not known. Certainly the immediate prognosis, with surgical treatment, is very good. In this series there were no operative deaths. All the cases showed marked and sustained improvement following surgery. Five of the patients went on to make excellent recoveries and were left with either no significant or only minimal disability. The 2 male patients are working full time at their original occupations. The 5 female patients, now all married, are performing normal household duties. Two, however (*Cases* 2 and 3), who have a moderately severe residual hemiparesis with sensory loss require some assistance.

Up to the present time no patient in this series has had a recurrence of bleeding. If, in fact, the bleeding in cases of subcortical hæmorrhage in young adults does occur from very small angiomatous malformations, and these are destroyed at the time of rupture, then the prognosis should be much better than in cases of cerebral hæmorrhage from other causes.

SUMMARY

1. Seven cases of subcortical hæmatoma treated by surgery are reported.

2. The possible role of small angiomatous malformations in the aetiology is discussed.

3. Comment on the clinical picture and diagnostic procedures is given.

4. The operative procedures and prognosis are briefly discussed.

I would like to thank Mr. Charles Langmaid and Mr. Norman Whalley for permission to publish these cases, and also for helpful criticism in the preparation of this paper. My thanks are also due to the Photographic Department of the Cardiff Royal Infirmary for the photographic reproductions.

Addendum.—Since the completion of this paper an important contribution has been made by Crawford and Russell (1956) on the role of small vascular malformations in the production of spontaneous intracerebral hæmorrhage.

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ENDOMETRIOMA OF THE RECTUM

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IN 1860 Rokitansky first described adenomyomata of the uterus. Since then numerous reports have been published of ectopic endometrial growth or implants in various pelvic and abdominal organs.

FIG. 713.—Barium enema examination. Lateral view to show the small filling defect at the pelvirectal junction.

Endometrial deposits in the large bowel are occasionally seen as an accompaniment of widespread endometriosis. Isolated endometriomata have also been reported, and their differentiation from carcinoma of the large bowel has been the subject of discussion by many authors. The occasional association of carcinoma of the bowel and endometriosis has also been described. The case reported here illustrates the difficulty that may be experienced in diagnosis both before and at the time of operation.

CASE REPORT

Mrs. C. D., aged 52, had complained of tenesmus for one year prior to her admission to the Whittington Hospital on Feb. 7, 1956. The tenesmus recurred at regular monthly intervals commencing the day before

FIG. 714.—Deep furrow in serosal aspect of lower end of specimen resembling a 'string' carcinoma.

menstruation and disappearing with the onset of the menstrual flow. During the month immediately preceding admission, however, the rectal discomfort had been continuous. The fæces had contained a little mucus from time to time, but there was nothing else abnormal about them. There had been no rectal bleeding.

Menstruation was regular, recurring every 28 days and lasting for 2–3 days. For several months she had had attacks of hypogastric pain at the onset of menstruation and this had tended to pass into the left iliac fossa.

This pain had been continuous for one month before admission.

In 1930 she had had an attack of acute nephritis, and hypertension was first noted in 1950.

ON EXAMINATION.—The patient was of average build with slight puffiness around the eyes. Apart from a blood-pressure of 176/116 mm. Hg, the cardiovascular and respiratory systems were normal.

FIG. 715.—The rectum opened to show the mass obstructing the lumen.

FIG. 717.—Section of the lesion to show several islands of endometrial tissue beneath the rectal mucosa. (H. and E.) (× 3.)

Abdominal examination revealed no obvious abnormality. On vaginal and rectal examination an irregular mass was felt in the pouch of Douglas and left lateral fornix. The uterus was axial, firm, irregular, and fixed.

Sigmoidoscopy was carried out to 18 cm. A good view was obtained and no abnormality of the rectum seen. Further instrumentation was prevented by pain on trying to negotiate a flexure of the bowel at this level.

A barium enema was reported as normal, the small filling defect at the pelvirectal junction being considered to be due to the normal narrowing so often seen at this level, and well shown in *Fig.* 713. In retrospect we feel that this radiological finding is, in fact, due to the lesion subsequently found at operation.

AT OPERATION (Feb. 10, 1956).—The abdomen was opened through a mid-line sub-umbilical incision.

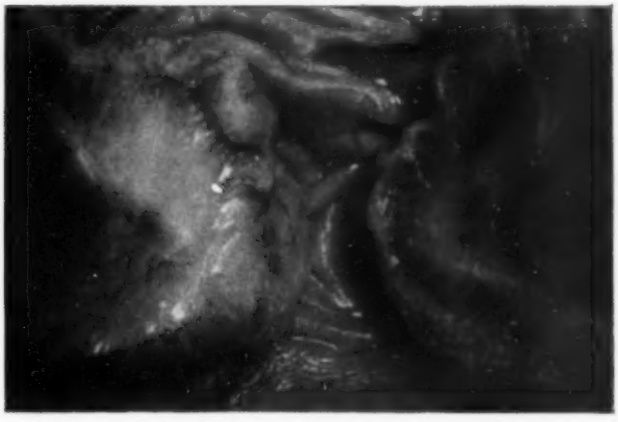

FIG. 716.—Enlarged view of obstruction to show intact rectal mucosa.

The uterus was found to be slightly enlarged by several small subserous and intramural fibroids. The posterior surface was adherent to the pelvirectal junction by what appeared to be endometriosis. Separation of uterus and large bowel was easy and revealed a hard tumour 4 cm. in diameter in the bowel wall at the pelvirectal junction. The bowel was narrowed at this point and the serosal surface was puckered. The appearance was typical of a 'string' carcinoma. There were no secondaries in the peritoneum, liver, or para-aortic lymph-nodes.

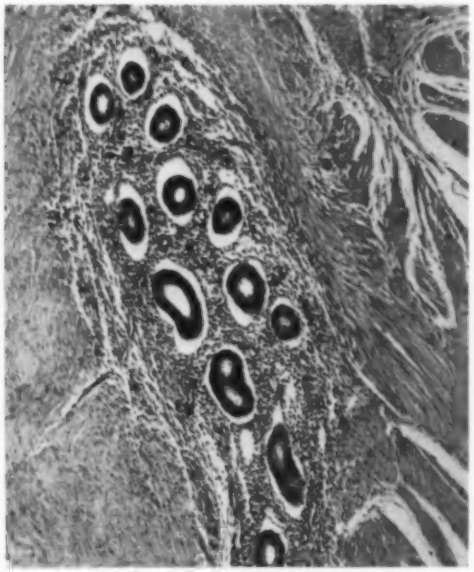

FIG. 718.—Enlarged view of an island of endometrial glands and stroma beneath the muscularis. (× 40.)

The rectum and pelvic colon were mobilized from the posterior abdominal wall and the hollow of the sacrum,

and the inferior mesenteric vessels were divided just below the left colic artery. The rectum was divided 5 cm. below the lower border of the lesion and the pelvic colon was divided 12·5 cm. above the growth.

The continuity of the bowel was restored by an end-to-end anastomosis. The pelvic peritoneum was reconstituted. The uterus and appendages were conserved. The abdomen was closed in layers and without drainage. Convalescence was completely uneventful.

Sigmoidoscopy was carried out on the 17th post-operative day and silk stitches of the suture line were seen at 15 cm. There was no stenosis of the bowel.

PATHOLOGY.—The resected specimen (*Fig.* 714) consists of a portion of colon 17 cm. long by 4 cm. diameter; 4 cm. from the lower end there is a deep furrow, 3 cm. long, on the peritoneal surface, running across the antimesenteric border.

The invaginated peritoneum shows marked fibrous thickening, in which there are a few conspicuous brown dots up to 0·1 cm. diameter; beneath this the muscle coat is hypertrophied, up to 2 cm. thick, and forms a hemispherical mass covered by intact folded grey mucosa, projecting 3 cm. into the lumen (*Figs.* 715, 716).

Histological examination shows great hypertrophy of the muscularis and severe fibrosis of the serosa. Numerous islands of endometrium consisting of abundant tubules lined by tall columnar epithelium surrounded by dense stroma are present. They lie mainly between the muscularis and the serosa, but there are a few tubules in the muscular and submucous coats and on the serosal surface. The mucosa is normal (*Figs.* 717, 718).

DISCUSSION

Green-Armytage (1933) reported an incidence of endometriosis in 8·9 per cent of 1000 abdominal operations performed for a variety of causes. Cattell (1937) in a series of 104 cases of endometriosis had an incidence of 16·3 per cent of rectal, sigmoid, or colon involvement. Sutler (1947) and Wilson and Tyson (1954) estimated that between 8 per cent and 15 per cent of all women have endometriosis during active menstrual life, and that of these about 25 per cent have involvement of the rectosigmoid. Approximately 2–4 per cent have rectosigmoid endometrial invasion of some degree. In the series reported by Kelly and Schladman (1949), 18 per cent of their cases of endometriosis had bowel involvement. Colcock and Lamphier (1950) in a series of 307 cases of endometriosis found an incidence of 18·3 per cent of bowel involvement, and of these, 40 per cent showed narrowing or obstruction of the lumen.

It can be seen from these reports that endometriosis of some degree is a common condition in women and that not a few of these have involvement of the large bowel. It is also apparent that other disease of the bowel, e.g., carcinoma, must occasionally be accompanied by endometriosis.

Endometrioma of the rectum can often be diagnosed before operation in the child-bearing period of life, but in later years considerable difficulty may be found in differentiating it from a carcinoma and even the operative findings may show a marked similarity between the two conditions.

Thierstein and Allen (1946) have listed the symptoms of rectal endometriomata in order of the frequency of their occurrence as follows :—

Rectal pain or pain on bowel movement; dyspareunia; backache; constipation; pain radiating down the thighs; menorrhagia; diarrhœa; nausea or vomiting; fainting spells; and blood in the stools. They emphasize that all of these symptoms are more pronounced during menstrual life.

Maclean (1936) and Mayo and Miller (1940) have noted the intensification of symptoms during the menstrual periods. In discussing the differential diagnosis of carcinoma and endometrioma the latter authors have also noted the different age groups, 87 per cent of endometriomata occurring between the ages of 30 and 49. Relative sterility is more common in cases of endometriosis, and in this condition the symptoms are present for a longer period than those commonly associated with carcinoma and inflammatory conditions. The symptomatology of carcinoma, however, may differ in no way from that of endometrioma in some instances, and where a carcinoma coexists with endometriosis an exacerbation of symptoms at the period time would be expected.

Sigmoidoscopy may well be inconclusive in the differential diagnosis. If the bowel lesion is more than 25–30 cm. above the external sphincter then inspection of the mucosa may be impossible. Although ulceration of the mucosa is uncommon in endometriosis its appearance may closely mimic that seen in an ulcerating carcinoma.

Barium enema examinations are often difficult to interpret. Maclean (1936) considered that it contributed little evidence to the diagnosis of the tumour and that there were no characteristic filling defects. More recently, however, some typical changes have been described. Josefsson (1939) emphasized the importance of demonstrating a normal mucosal pattern at the site of a filling defect in endometriosis and suggested the value of radiographic study of a questionable lesion at the time of menstruation, when stenosis of the bowel may appear more marked than in the interval between the periods. Jenkinson and Brown (1943) have also described certain radiographic changes which they believe to be typical. These are relatively long filling defects of 10–18 cm., a sharp demarcation of the filling defects with other portions of the bowel showing little evidence of the disease, an intact mucosa, and exquisite tenderness on palpation during screening.

Whilst accepting these observations, we were unable to find the 'typical changes' in the case reported here and as already stated, the small defect seen was identical with similar defects seen in a normal bowel.

The findings at operation may, again, be deceptive. Thierstein and Allen (1946) described a superficial and deep type of endometrioma. The former was characterized by a puckering in the serosa and a hard nodule varying from a buckshot to a bean size. The latter consisted of larger masses mainly situated in the muscularis. Other authors, notably Maclean (1936) and Macleod (1947), believe that the appearances presented may resemble those of a scirrhous carcinoma. Maclean believed, however, that there were three significant points of difference to enable one to distinguish carcinoma from endometrioma.

1. The stricture does not tend to encircle in endometrioma.

2. Endometrioma produces a puckering in the serosa rather than the appearance of a string tied

around the gut which is so characteristic of scirrhous carcinoma.

3. An endometrioma can be lifted up as a button in the bowel wall and moved without moving the whole segment of colon or rectum.

Maclean appreciated, however, that all these features might be presented by an early carcinoma.

Macleod (1947), in his classical paper, believed that in many cases differentiation was not possible and in all cases in which there is doubt resection of the affected loop is the proper course. Smith (1943) had previously voiced the same opinion.

Whether a frozen section should be examined at the time of the laparotomy is a matter of some conjecture. Colcock and Lamphier (1950) were emphatic that this should be done. We felt that there was some danger in taking a biopsy of the tumour from the serosal surface, and that the clinical findings justified an immediate anterior restorative resection.

All authors writing on this subject are agreed upon the treatment of endometrioma of the large bowel once a diagnosis has been made, and this can be summarized thus :—

1. When preservation of fertility is important, resection of the mass, if it is causing obstruction, with an end-to-end anastomosis should be performed wherever possible.

2. When preservation of fertility is no longer important and the patient is over 45, a hysterectomy and bilateral salpingo-oöphorectomy should be carried out. This procedure has been noted to reduce very large masses and to restore completely the patency of the bowel (Grave, 1927 ; Hutchins, 1935 ; Friedman, 1936 ; Cattell, 1937 ; Schofield and Bacon, 1938 ; Patton and Patton, 1941 ; Jenkinson and Brown, 1943 ; Smith, 1943 ; Ben-Asher, 1944 ; Miller, 1944 ; Macleod, 1947 ; Owen and Stone, 1949 ; Marshak and Friedman, 1950 ; Hawthorne, Kimbrough, and Davis, 1951 ; Benz, Dockerty, and Dixon, 1952).

The case reported here presented many features of a carcinoma and but few of an endometrioma. The appearances at operation were as typical of a carcinoma as of an endometrioma and the endometriosis seen at operation was not extensive. We felt justified, therefore, in performing an anterior restorative resection, but we agree that one criticism of the treatment could be the failure to ablate the ovaries in the presence of some endometriosis. However, the diagnosis of carcinoma had been confidently made and the remaining endometriosis was not widespread.

SUMMARY

A case of endometriosis with involvement of the rectosigmoid has been described.

The differential diagnosis and the pitfalls therein have been discussed.

The treatment of cases of endometrioma of the rectum and rectosigmoid have been outlined.

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SEPSIS IN SURGICAL WOUNDS WITH PARTICULAR REFERENCE TO STAPHYLOCOCCUS AUREUS*

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In spite of antibiotics sepsis in surgical wounds, especially with antibiotic-resistant *Staphylococcus aureus*, is still a major problem (Howe, 1954).

During a survey of staphylococcal cross-infection in surgical wards in this hospital, it was found that 80 per cent of the 154 moist wounds in one of the wards were infected at some time with *Staph. aureus*, and that 31 per cent of patients with drained wounds went home with a wound still discharging *Staph. aureus*. In the present paper the severity of the problem of staphylococcal sepsis as seen during that survey is assessed. Throughout the paper *Staph. aureus* refers to a coagulase-positive staphylococcus and *Staph. albus* refers to a coagulase-negative staphylococcus (*Staph. saprophyticus*, Shaw, Stitt, and Cowan, 1951).

MATERIAL AND METHODS

The investigation was carried out during ten months in two general surgical wards (Clarke, Dalgleish, Parry, and Gillespie, 1954). All except hæmorrhoidectomy wounds were swabbed with moistened swabs every time they were dressed, except for those dressed daily which were swabbed about every four days. At the time of swabbing a note was made of the appearance of the wound. Bacteriological methods used were as described by Clarke and others (1954).

The part of the investigation described below concerns only dry wounds and drained wounds. Suprapubic and colostomy wounds are not considered. Dry wounds can be defined as undrained wounds which normally healed by first intention. They rarely became infected. Drained wounds often became infected, but since the drain usually caused mechanical inflammation and organisms may have grown saprophytically in the discharge, it was often difficult to say whether the organisms isolated were causing inflammation or not.

RESULTS

The Cause of the Break-down of Dry Wounds.—Out of the 382 dry operation wounds investigated, 52 (13·6 per cent) broke down; that is, they healed by second intention. These included wounds from operations such as appendicectomy and partial gastrectomy involving the opening of unsterile viscera. In 27 (7·1 per cent) the break-down was minimal, but in 25 (6·5 per cent) it was moderate to severe (*Table I*).

It will be seen that the most important cause of serious break-down was the bursting of abdominal abscesses which yielded coliform bacilli on culture. The majority of these occurred after appendicectomy and were probably due either to the contamination of the layers of the abdominal wall during the operation or to the formation, after peritonitis, of intra-peritoneal abscesses which subsequently burst outwards.

The second most important cause of the serious break-down of clean wounds was infection by *Staph. aureus*. It appeared that of strains of *Staph. aureus* causing break-downs, all but one were acquired in the theatre. This was thought to be so because the wounds were either found to be discharging copious pus at the time of the first dressing or, more often, they were apparently healing well until the fifth to ninth days after operation, when a deep subcutaneous abscess appeared which later burst.

Table I.—The Causes of the Breaking-down of Dry Wounds

Cause of Wound Break-down	Degree of Sepsis			
	Minor	Moderate or Severe	All Types No.	All Types Per cent
Staph. aureus infection :—				
Penicillin-resistant	3	3	6	11·5
Penicillin-sensitive	3	4	7	13·4
β-hæmolytic streptococcus infection	0	1	1	1·9
Staph. albus infection	15	3	18	34·6
Anærobic cellulitis after abdominal operation	0	1	1	1·9
Bursting of deep coliform abscess	3	10	13	25·0
Gaping of incision after removal of clips	1	2	3	5·8
Mechanical effect and *Staph. albus* infection	2	0	2	3·8
No information	0	1	1	1·9
Total	27	25	52	

The total break-down rate found here (13·6 per cent) is higher than that found by other workers for clean wounds (Eliason and McLaughlin, 1934; Meleney, 1935; Ives and Hirshfeld, 1938; Devenish and Miles, 1939; Logue and McKissock, 1945). However, the figure includes many wounds with trivial break-downs. If only moderately and severely septic wounds are considered, the rate of 6·5 per cent is in the upper range of the rate found by others.

It is of interest that half of the wound abscesses due to *Staph. aureus* acquired in the theatre yielded penicillin-sensitive staphylococci; whereas it has been found that coagulase-positive staphylococci acquired in the ward or found in the noses of nurses were nearly always penicillin-resistant. The sensitive staphylococci may have come from the patients' own noses and skins, since operations were performed usually soon after admission. Of the 7 patients with abscesses due to penicillin-sensitive *Staph. aureus*, 3 were nasal carriers of penicillin-sensitive *Staph. aureus* at the time of operation. On the other hand, the result might have been because the wounds were infected by the hands of surgeons who were nasal or skin carriers of penicillin-sensitive

* This paper is based on part of a thesis for the degree of M.D. in the University of Bristol.

Staph. aureus. Doctors and students do not acquire penicillin-resistant staphylococci as often as do nurses (Barber, Wilson, Rippon, and Williams, 1953), and possibly consultant surgeons acquire them less often than do hospital residents. The result indicates that infection in the operating theatre probably did not originate directly or indirectly from nurses in half the cases.

The Causes of Minor Inflammation in Dry Wounds.—As well as the minor break-downs shown in *Table I*, many dry wounds not infected with organisms usually accepted as pathogens were slightly red or gaping. To see whether either this minor inflammation or the minor break-down discussed above was due to the actual process of healing, to the mechanical irritation of the sutures, or to infection with *Staph. albus* or diphtheroids, which are usually considered to be commensals, the clinical state, the result of swabbing, and the suture material of all dry wounds which were not infected with pathogenic organisms either at the time of swabbing or previously, were investigated (Clarke, 1955). If a wound was swabbed several times, one swab was counted for each of the periods 3–5, 6–8, and 9–14 days after operation. One wound might therefore be counted up to three times in the 3–14 day totals. Any difference between the different groups studied below might therefore be exaggerated among this set of totals. Almost all the wounds were closed with silkworm gut sutures of average thickness or with Michel clips.

1. *Inflammation in Relation to Suture Material.*—*Table II* shows that statistically significantly more wounds closed with stitches only were inflamed, than those closed with both stitches and clips (there were usually one or two stitches and many clips in these wounds), and statistically significantly more of these were inflamed than those closed with clips only ($\chi^2 = 19{\cdot}7$, $p < 0{\cdot}01$). Since the maximum incidence of inflammation was 6–8 days after operation and since the numbers of colonies of *Staph. albus* isolated from the wound are shown below to have affected the incidence of inflammation, a true relationship can be shown only by considering separately, for example, wounds swabbed 3–5 days after operation, and at the same time yielding between 10 and 50 colonies of *Staph. albus*. These wounds showed a similar difference between stitches and clips, but the total numbers are too small to calculate significance. Almost all the wounds studied were abdominal, and the distribution of the different types of operation was much the same in the groups of wounds closed with different materials.

The increased tendency to inflammation among wounds closed with stitches is consequently probably real. Stitches therefore predisposed to minor inflammation in wounds, but whether this was due to purely mechanical irritation, or to mechanical irritation allowing *Staph. albus* to gain entrance to the wound cannot be decided. Clips accordingly should cause less discomfort than stitches and lead to less serious secondary sepsis. On the other hand, after clips were removed the wound occasionally gaped and was therefore slow in healing and sometimes became severely secondarily septic. The important fact may be that clips were removed on the fourth day and stitches after the sixth day. Perhaps if stitches were removed on the fourth day they would cause

Table II.—INFLAMMATION IN DRY WOUNDS IN RELATION TO INFECTION WITH *Staph. Albus*, TYPE OF CLOSURE, AND THE LENGTH OF TIME AFTER OPERATION

No. of *Staph. albus* isolated	Days after operation	Stitches Only			Stitches and Clips			Clips Only			Total		
		No. of wounds	No. inflamed	Percentage inflamed	No. of wounds	No. inflamed	Percentage inflamed	No. of wounds	No. inflamed	Percentage inflamed	No. of wounds	No. inflamed	Percentage inflamed
More than 50	3- 5	16	6	37	15	2		15	1		46	9	19·6
	6- 8	42	28	66·7	6	6		4	2		52	36	69·3
	9-14	14	4	29	9	7		1	0		24	11	46
	3-14	72	38	52·7	30	15	50·0	20	3	15	122	56	45·9
10-50	3- 5	14	5	36	11	1	9	20	4	20	45	10	22·2
	6- 8	20	7	35	14	6		4	1		38	14	36·8
	9-14	7	5		11	5		0	0		18	10	56
	3-14	41	17	41·5	36	12	33·4	24	5	21	101	34	33·7
1-10	3- 5	11	4		10	2		20	2		41	8	19·5
	6- 8	16	7	44	10	4		5	2		31	13	42·0
	9-14	15	4		8	2		0	0		23	6	26
	3-14	42	15	35·7	28	8	28·5	25	4	16	95	27	28·4
None	3- 5	11	3		24	5		12	0		47	8	17·0
	6- 8	16	6	38	8	0		1	0		25	6	24
	9-14	12	4		9	2		1	1		22	7	32
	3-14	39	13	33·4	41	7	17·0	14	1	7	94	21	22·3
Total	3- 5	52	18	34·6	60	10	16·7	67	7	10·4	179	35	19·6
	6- 8	94	48	51·0	38	16	42·2	14	5	36	146	69	47·3
	9-14	48	17	35·4	37	16	43·3	2	1		87	34	39·1
	3-14	194	83	42·8	135	42	31·1	83	13	15·7	412	138	33·5

Percentages calculated from a denominator of less than 25 are shown to the nearest whole number, and percentages calculated from a denominator of less than 10 have been omitted.

less inflammation. Perhaps stitches may sometimes have been too tight and reduced the vitality of the tissues. On the other hand, the important factor may be that clips are not pushed deeply through the skin. When stitches are inserted, *Staph. albus* in the deeper layers of the skin, having survived the antiseptic applied to the skin surface, may be pushed along the suture track and there give rise to inflammation. Gillman, Penn, Bronks, and Roux (1955) have shown that there are many histological changes in the suture track.

2. *Inflammation in Relation to the Numbers of Staph. albus.*—*Table II* shows that for all wounds together there was a statistically significant correlation between the amount of growth of *Staph. albus* and the proportion of wounds which were inflamed ($\chi^2 = 14{\cdot}84$, $p < 0{\cdot}01$). There was no such correlation for wounds swabbed 3–5 days after operation or for wounds closed with clips only. The correlation was, however, demonstrable and statistically significant when wounds closed with stitches only, and at the same time swabbed 6–8 days after operation were considered separately ($\chi^2 = 6{\cdot}31$, $0{\cdot}01 < p < 0{\cdot}02$). The correlation must therefore be considered real and not due to any differences in distribution as regards day of swabbing or the type of closure employed.

This result seems to indicate that *Staph. albus* often causes mild inflammation in dry wounds. This was considered to be possible by Meleney (1941), Hare and Willits (1942), and Logue and McKissock (1945). On the other hand, the inflammation might have been due to mechanical causes, such as the irritation caused by the stitches, and the *Staph. albus* might have grown saprophytically in the discharge (Williams and Miles, 1949). The fact that there was no correlation between *Staph. albus* and inflammation in wounds closed with clips supports this theory. The correlation is demonstrable when wounds closed with stitches only are considered alone. It is possible that in those wounds closed by stitches only which showed inflammation, the stitches were causing more irritation than in those which were not inflamed, perhaps because of variations in reaction to stitches in different people. The most probable explanation is that *Staph. albus* played some part in causing minor inflammation. This conclusion is strengthened by the finding that there was no statistical correlation between the growth of diphtheroids from a wound and the incidence of inflammation.

The Severity of Infections caused by Penicillin-resistant and Penicillin-sensitive *Staph. aureus.*—This was investigated because of reports that penicillin-resistant *Staph. aureus* was possibly less virulent than penicillin-sensitive *Staph. aureus* (Sherris and Florey, 1951).

The notes were studied of all patients whose wounds had yielded *Staph. aureus* at any time (Clarke, 1955). Of those that were inflamed, those where the *Staph. aureus* was not the only possible cause of the inflammation were excluded. There remained 31 wounds infected with penicillin-sensitive *Staph. aureus* and 76 infected with penicillin-resistant *Staph. aureus* to be studied. In each case the amount of inflammation in the wound, the presence of pyrexia over 99° F., and the length of time the wound discharged pus, were recorded. Sometimes the patients' notes did not yield enough information on one of these points. In that case the wound was recorded under that point as "not known". Pyrexia of any duration occurring at the same time that the wound was infected was recorded only if no cause for it, other than wound infection, could be found.

Table III shows that the majority of non-drained wounds infected with *Staph. aureus* did not become inflamed. There was no difference between the proportion of non-drained wounds infected with penicillin-resistant *Staph. aureus* and those infected with penicillin-sensitive *Staph. aureus* which became mildly or severely septic, or which discharged for more than 18 days, or which caused pyrexia.

For all wounds taken together the proportion which were mildly or severely septic was statistically significantly higher in the penicillin-resistant group than in the penicillin-sensitive group ($\chi^2 = 4{\cdot}08$, $0{\cdot}02 < p < 0{\cdot}05$). This may have been due partly to the fact that the penicillin-resistant group contained

Table III.—THE SEVERITY OF INFECTIONS CAUSED BY PENICILLIN-RESISTANT AND PENICILLIN-SENSITIVE *Staph. Aureus* IN DRAINED AND ORIGINALLY DRY WOUNDS

		PENICILLIN-SENSITIVE					PENICILLIN-RESISTANT				
		Not drained		Drained	Total wounds		Not drained		Drained	Total wounds	
		No.	Per cent	No.	No.	Per cent	No.	Per cent	No.	No.	Per cent
Inflammation	Moderate and severe	4	18	4	8	25·8	6	17·2	20	26	34·2
	Mild	1		1	2	6·5	1		13	14	18·4
	None	17		4	21	67·7	27		4	31	40·7
	Not known	0		0	0		1		4	5	
Pyrexia due to wound	Present	2	9	2	4	12·9	4	11·4	16	20	26·3
	Absent	20		7	27		31		25	56	
Wound discharge	More than 18 days	4	18	3	7	22·5	4	11·4	12	16	21·0
	Less than 18 days	18		5	23		28		23	51	
	Not known	0		1	1		3		6	9	
Total number of bacteriologically infected wounds		22	100·0	9	31	100·0	35	100·0	41	76	100·0

more patients who were severely ill and whose wounds were drained for a long time. These wounds would be more likely to become inflamed if they were infected with *Staph. aureus*. Also wounds which were infected with penicillin-sensitive *Staph. aureus* would respond better to treatment with antibiotics, which would prevent their becoming seriously inflamed.

On the other hand, once a patient's wound had become infected with a penicillin-resistant *Staph. aureus*, it was swabbed and its appearance noted only once a fortnight. Therefore the description of the wound was usually that made when the infection was just starting. It is probable that most of these infections subsequently became worse. The infections caused by penicillin-resistant *Staph. aureus* were in fact probably more severe than is shown in the table.

Table IV.—THE REASONS FOR PATIENTS WITH DRAINED WOUNDS STAYING IN HOSPITAL MORE THAN THE NORMAL LENGTH OF TIME

Reason for Staying in Hospital for More than Maximum Normal Time			Patients Staying in Hospital for More than Normal Time		Bed-days Wasted		Bed-days Wasted per Patient
			No.	Per cent	No.	Per cent	No.
Discharging wound	Sepsis	*Staph. aureus*	18	30·0	145	22·9	8·1
		Other organisms	5	8·3	36	5·7	7·2
	Biliary, fæcal, urinary fistula, or hæmatoma		9	15·0	135	21·3	15·0
Venous thrombosis or pulmonary embolism			2	3·3	19	3·0	9·5
General illness			23	38·4	250	39·4	10·9
Social reasons			3	5·0	49	7·7	16·3
Total			60	100·0	634	100·0	10·6

That strains of penicillin-resistant *Staph. aureus* were less virulent than strains of penicillin-sensitive *Staph. aureus* was not demonstrated. This confirms the work of Barber (1947) and of Reiss, Pulaski, Amspacher, and Contreras (1952). The result found by Sherris and Florey (1951), as they themselves suggested, must have been due to the fact that their two groups were not comparable.

Bed-wastage due to *Staph. aureus*.—The average length of stay in hospital of patients whose wounds were infected with *Staph. aureus* was found to be 5 days longer than the average length of stay of patients whose wounds were not so infected. However, many patients stayed in hospital for more than the usual length of time for reasons not connected with staphylococcal infection. These patients then, owing to their long stay in hospital, acquired *Staph. aureus* in their wounds.

Again, patients who had had certain types of operation which necessitated their staying in hospital for a long time had also the type of wound which often acquired *Staph. aureus*. Also, in the ward where fewer wounds became infected with *Staph. aureus* it was the custom to send the patients home in the shortest possible time after operation, unlike the custom in the other ward.

The notes of all patients with drained wounds were examined, and those who had died, or whose wounds were infected with tubercle bacilli, or whose wounds were large granulating spaces, were excluded. The maximum normal time and the average normal time from the day of operation till discharge were found for each type of operation on the patients who had apparently had no complications. The probable reason why each patient who developed complications stayed in for more than the maximum normal time was decided. The number of bed-days wasted by each patient was calculated as the number of days the patient stayed in hospital after operation in excess of the average normal stay of the uncomplicated patients.

Of the 135 patients with drained wounds, 60 (44·5 per cent) stayed in hospital for more than the maximum normal time. The chief reason for prolonged stay in hospital (*Table IV*) was, as might be expected, severe surgical illness. The second most common reason was wound sepsis due to *Staph. aureus*, for which reason, 18 (30 per cent) patients stayed in, causing a wastage of 145 bed-days. This meant that each patient wasted 8·1 bed-days, which is about the same wastage as found by Eliason and McLaughlin (1934). Goodall (1952) found a larger bed-wastage, but this was not all due to staphylococcal wound sepsis.

During the period of the investigation 71 bed-days were wasted by *Staph. aureus* causing sepsis in dry wounds (10 patients). Thus 216 bed-days were wasted during 8½ months in 2 surgical wards by sepsis due to *Staph. aureus* in drained and dry wounds. This works out as 1·3 per cent of all bed-days. This figure is a minimum estimate, for it does not include staphylococcal sepsis in other types of wound such as suprapubic wounds and colostomies, or other staphylococcal complications such as pneumonia and urinary infections, or staphylococcal sepsis in patients who died. Also there were limitations to the method of calculation. It was impossible to work out exactly how many bed-days were wasted by staphylococcal sepsis, for although it was known that some patients had stayed in hospital for more than the usual time because of wound sepsis, it was not known for exactly how long. There must have been other patients who stayed in hospital because of wound sepsis on whose notes the fact was not recorded.

At a very rough guess the true total wastage due to *Staph. aureus* was probably in the region of 300 bed-days (1·9 per cent of all bed-days). This figure is surprisingly low.

SUMMARY

Wound infection was studied in two general surgical wards over a period of 10 months; 13·6 per cent of clean, dry wounds broke down, 6·5 per cent of them seriously. The most common cause of severe break-down was the bursting of deep abdominal abscesses yielding coliform bacilli. The second most common cause was sepsis due to *Staph. aureus*. These infections were almost always acquired in the theatre and half were penicillin-sensitive.

Minor inflammation in healing dry wounds was commoner 6–8 days after operation than at other times; it was commoner in wounds closed with stitches only than in those closed with clips; and it was commoner in wounds heavily infected with *Staph. albus* than in those lightly infected or not infected.

Strains of penicillin-resistant *Staph. aureus* caused as severe lesions as strains of penicillin-sensitive *Staph. aureus*.

A rough estimate was made of the amount of bed-wastage due to *Staph. aureus*.

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MAFFUCCI'S SYNDROME

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THE association of dyschondroplasia and vascular hamartomata (cavernous angiomata with phlebectasia) is a rare disorder first described by Maffucci in 1881 (Carleton, Elkington, Greenfield, and Robb-Smith, 1942). In the skeleton there is a disturbance of bone formation in the cartilage at the growing ends of bone. Cartilage persists in the diaphysis and forms enchondromata and eccondromata, and the imperfect ossification and irregular growth may cause severe deformities. The lesions may affect any part of the skeleton, especially the long bones and phalanges. The skull, carpus, and tarsus are involved less frequently. Pathological fractures are common and delayed union and malunion are frequent. Malignant change occurs in the chondromata in about a quarter of the patients.

The vascular anomalies consist of multiple cavernous hæmangiomata and phlebectasia. They may occur anywhere in the subcutaneous tissue, and form reddish-blue, soft, and slightly tender tumours. These tumours may become calcified and are then visible on soft-tissue radiographs. Phlebectasia is common and is present in more than half of the patients. The vascular changes are associated with, but do not appear to determine, the skeletal lesions: the linkage can therefore be regarded as coincidental rather than consequential. The affected persons are generally of low stature and poor muscular development, but of average intelligence. They are normal at birth, and the angiomatous and skeletal changes always appear before puberty. The pathogenesis of the syndrome is uncertain, but it is probably a mesodermal dysplasia. A neurotrophic factor is suggested by the striking tendency to one-sidedness, associated in certain cases with facial asymmetry and vitiligo.

CASE REPORTS

Case 1.—A male aged 14 years, who was quite normal at birth, had, since the age of 8, frequent heavy nasal bleedings, and during the past five years had thrice fractured the left forearm after minor trauma. On each occasion the union was unusually slow. Four years ago his left ring finger became deformed, and numerous subcutaneous hæmangiomata appeared along the finger. The hæmangiomata continued to arise almost entirely on the left side of the body, and especially on the foot where some had purplish centres and were 1–2 cm. in diameter.

The skeletal lesions were not severe and were confined to the left side of the body: chondromata were present in the phalanges and the metacarpals of the left hand (*Fig.* 719), in the lower end of the left ulna, and in the mid-shaft of the left humerus. The right hand and

upper limb bones were normal. Chondromata were also present in two of the left metatarsals, and in the lower half of the left tibia. The two opacities posterior to the left talus are calcified phleboliths (*Fig.* 720). There was no abnormality in the skull or spine apart from a slight alteration of the trabecular pattern of the lower lumbar vertebræ.

Histological examination of a subcutaneous nodule from the left leg was reported as a thin-walled cavernous angioma (*Fig.* 721), and of the tumour of the third left metacarpal a simple enchondroma. The skin temperature on the left side was slightly raised as compared with the right. The blood-pressure was the same on both sides. On the electrocardiograph there was inversion of the T-wave in Leads II and III suggestive of myocardial damage. There was a marked leucocytosis and the blood-urea was slightly raised, otherwise the biochemical examination was negative. There was no abnormality in the cerebrospinal fluid.

FIG. 719.—Radiograph showing the skeletal changes in the left ulna, metacarpals and phalanges; while the right side is normal.

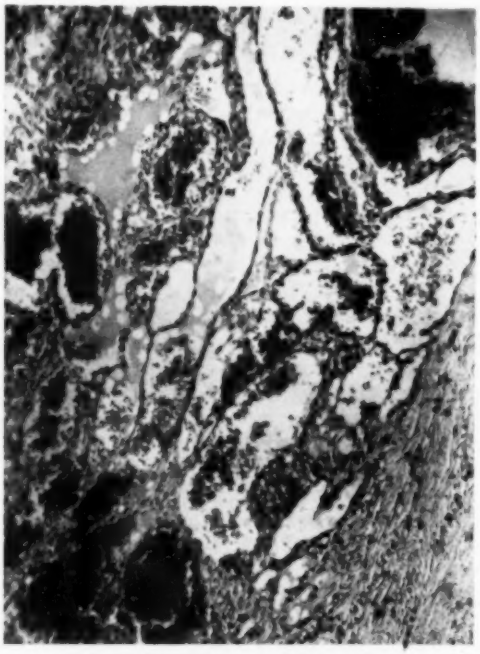

FIG. 721.—Photomicrograph showing the characteristically thin-walled angioma. H. and E. (× 140.)

Case 2.—A 13-year-old girl, first seen in 1949 when 7 years old, had a hæmangioma of the pulp of her left hallux (*Fig.* 722), which had been present since infancy. Radiographic examination revealed many exostoses arising from the metatarsals and phalanges of the left foot: the metatarsals were short and some of them showed translucent areas suggestive of chondromata (*Fig.* 723). It was noted that she had many exostoses

FIG. 720.—Radiograph showing two calcified phleboliths posterior to the left talus.

FIG. 722.—Photograph showing the shortened hallux with the hæmangiomata in its pulp.

elsewhere in the skeleton but none of these was painful. During 1952 she developed a bilateral genu valgum deformity due to growth disturbance at the upper ends of both tibia. The deformity was corrected by a high right and left tibial osteotomy, and the exostoses at this site were excised. A year later the genu valgum had recurred, and the medial side of both lower femoral

FIG. 723.—Radiograph of feet with multiple enchondromata in the tarsal and metatarsal bones.

epiphyses were stapled and the exostoses excised at this site (*Fig.* 724). After this operation the genu valgum steadily improved and there is now no appreciable deformity. The electrocardiograph illustrates a flat T-wave in Lead III. As in the first case there was a mild polymorphonuclear leucocytosis, but the biochemical examinations of the blood and cerebrospinal fluid were normal. The skin temperature on the left foot was 26·6° C. and on the right foot 23·6° C. The blood-pressure was also raised on the left arm and leg, 130/90 mm. Hg, as compared with 120/70 mm. Hg on the right side.

Case 3.—A 16-year-old youth of normal stature who, since early infancy, has had on the anterolateral aspect of his right leg cavernous hæmangiomata, but no exostoses. Six years ago he noticed that the right leg was longer, thicker, and warmer than the left, most of the overgrowth being in the tibia.

The measurements were as follows :—

	Right in.	*Left* in.
At 3 in. below the tibial tubercle (diameter)	15½	14½
At 6 in. above the pole of patella (diameter)	19½	19½
Total length of the leg	40	38½
Length of the tibia	20½	19½

Here also there was a mild leucocytosis, but no abnormal biochemical findings in the blood or cerebrospinal fluid. The temperature of the right leg was 35° C., and that of the left 29° C. The blood-pressure in the right arm and leg was 130/80 mm. Hg, and on the left side 120/70 mm. Hg. The electrocardiograph revealed right ventricular preponderance with depressed T-waves in Lead III. Radiographic examination revealed no bone lesion in the legs or feet, but there was some calcification in the soft tissues around the mid-shaft of the left femur consistent with phleboliths.

It is interesting that his younger brother suffers from multiple diaphysial aclasis, but no vascular lesions are present. This might suggest that there is a genetic link between the skeletal and vascular

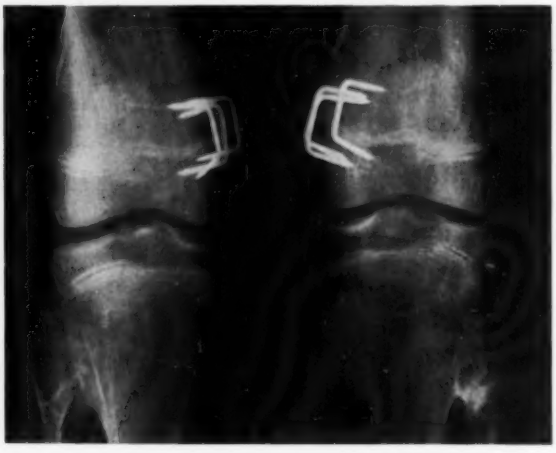

FIG. 724.—Radiograph showing stapling of the medial side of the lower femoral epiphyses in both knees.

manifestations of the syndrome; which could be due to the crossing over between two loci on a chromosome pair rather far apart.

SUMMARY

1. Maffucci's Syndrome consists of dyschondroplasia (Ollier's disease) in association with vascular hamartomata (cavernous angiomata with phlebectasia). Twenty-six cases have hitherto been described.

2. A further two cases are recorded both of which show either inversion or flattening of the T-wave in Lead III in the electrocardiographs.

3. *Case* 2 shows the complication of genu valgum and the difficulties in her treatment.

4. A genetic factor is suggested by a third patient who had the vascular anomalies of the syndrome without skeletal deformity, whilst in his brother all the skeletal changes were present, but no vascular changes.

Thanks are due to Mr. Roland Barnes and Professor D. F. Cappell for their advice and interest in this work. To Mr. Russell Thomson, and Dr. W. A. Taylor, for their assistance in *Case* 1.

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PROSTATIC ABSCESS

By KEITH CHITTY, Bristol

PYOGENIC abscess of the prostate, although a rare surgical condition, occurs sufficiently often in a urological practice for its diagnosis and treatment to provide an important problem. Several series of cases have been reported in the literature (Forsythe, 1942; Schwartz, 1940; Lowsley, 1939; Sargent and Irwin, 1931; Kickham, 1938; Bell, 1938). These have mostly been of small numbers, but Sargent and Irwin report 42 cases and Lowsley 59 cases.

The route of infection is generally recognized as being either direct from posterior urethritis, or metastatic from infections in other parts of the body. Views on the relative preponderance of these two routes have changed progressively with the years, since public health measures and sulphonamide and antibiotic therapy have reduced the incidence of chronic gonorrhœa. Thus Sargent and Irwin (1931) state that in their opinion 75 per cent of their cases were gonococcal in origin and only 4 of 42 cases were definitely metastatic, while Lowsley (1939) puts more emphasis on the metastatic source. More recently Forsythe (1942) thinks that gonorrhœa is now an unimportant cause and that most cases arising from a urethritis do so as a result of urethral instrumentation. A case such as that reported by Dunlop and Hosford (1952) caused by neglected gonorrhœa must therefore be considered a rarity. Accordingly metastatic infection assumes a greater importance and a variety of causative organisms have been described. The infection commonly results from staphylococcal skin infection, as does a carbuncle of the kidney, with which it may be coincident (Kickham, 1936), but may follow other suppurative processes such as pneumonia, appendicitis, or tonsillitis, with corresponding organisms. Coliform abscesses are also commonly found, but the source of this infection is less certain as the organisms are frequently found in the urinary tract, but in the absence of preceding urinary infection or instrumentation a metastatic origin is most likely.

The earlier writers give the age incidence as widely spaced—Lowsley (1939), for example, from 17 to 80 years—but emphasize the young age of its maximum incidence (20 to 40 years), and this is in keeping with the high incidence of gonococcal infection among their cases. Later writers, in smaller series, find a slightly higher average age. All agree that it is not a condition primarily associated with prostatic hyperplasia.

The symptoms are well described in the literature (e.g., Forsythe, 1942; Lazarus and Rosenthal, 1936) and consist of frequency and dysuria, difficulty of micturition or retention of urine, and pain felt in the perineum, rectum, or suprapubic region. It is recognized, however, that the early symptoms may be entirely constitutional with little relation to the lower urinary tract (Kickham, 1938). The diagnosis is made on the history combined with the physical signs, of which rectal examination is the essential part. This reveals a tender, hot, œdematous prostate, frequently asymmetrical, from which fluctuation may sometimes be elicited. This sign of fluctuation, however, is difficult to determine with certainty, and Sargent and Irwin (1931) in their series found it present only in one quarter of their cases. The differential diagnosis is primarily from acute prostatitis, from which it differs only in degree, and rests on the presence of this fluctuation or the demonstration of pus by aspiration or surgical drainage. Abscess of the seminal vesicle is usually associated with acute prostatitis and is particularly difficult to differentiate, except by the position of the fluctuant area above the tender inflamed prostate rather than centrally. In practice, differentiation also from adenoma, breaking down carcinoma and tuberculous abscess is not a simple matter. Methods of investigation are limited. A leucocyte count may help, and urethrography is advocated by Lowsley and by Forsythe as showing distortion of the urethra by unilateral swelling. Cystoscopy (Bell, 1938; Thompson, 1937; Timberlake, 1938) may be used as a more direct way of obtaining this information.

Various methods of treatment have been practised. Conservative treatment consists of local heat, rest, and medication with urinary antiseptics, systemic chemotherapy, or antibiotics in the expectation of resolution or natural discharge into the urethra, but rupture may occur instead into the rectum, perineum, bladder, perivesical tissues, or peritoneum with serious results, although exceptionally spontaneous cure may result. In all cases there is an inherent danger of incomplete drainage and of a residual chronic prostatitis developing, as emphasized by Thompson (1937).

Surgical intervention is usually indicated when abscess formation is established and many methods of drainage have been described and variously advocated or condemned by different authors (Sargent and Irwin, 1931; Lowsley, 1939; Millin, 1947; Timberlake, 1938; Bell, 1938; Wilhelm, 1945). These may be summarized as follows: (1) Blind rupture with a urethral sound; (2) perineal urethrotomy and rupture with a finger introduced into the prostatic urethra; (3) unilateral perineal incision and opening the abscess by Hilton's method; (4) perineal exposure and incision of the prostate under vision; (5) perineal prostatectomy; (6) a transvesical or retropubic approach; (7) endoscopic incision with a Colling's knife or special electroprostatome; (8) transurethral resection.

All these methods have been applied successfully to individual cases, but have also been criticized by other authors in the following terms. Blind rupture is an uncertain method which may result in imperfect drainage. Urethrotomy gives certain drainage, but involves a perineal incision with danger of delayed healing and damage to the urethral sphincter. Unilateral perineal incision with Hilton's method of drainage requires certain localization and will fail to drain an abscess present in the opposite lobe. Perineal exposure of the prostate gives good surgical

drainage, but involves extensive dissection through normal tissue planes; this is the method advocated in the recent British text-books (Winsbury-White, 1948; Riches, 1950; Badenoch, 1953). Trans-urethral methods are traditionally feared in the presence of acute inflammation as potential causes of ascending infection or septicæmia, but in practice no such sequels have been encountered even in the days before antibiotics. Schwartz (1940) reported a case unintentionally treated in this way when mistaken for carcinoma, and Bell (1938) reported 4 cases with no constitutional disturbance and a rapid convalescence.

The present series consists of 27 cases of prostatic abscess seen and treated in the genito-urinary department at Bristol during the past 6 years. This includes 1 patient who was admitted again after a 4-year interval with a second acute abscess. Thirteen were direct admissions, 9 were seen in the out-patient department, and 4 were referred by other surgeons. The age incidence is from 26 to 85 years, with an average of 56 years, which is rather higher than in most series. There is, therefore, a larger proportion falling into the ' prostatic age-group '. Nevertheless, in only 6 was there clinical evidence of hyperplasia and in only 4 associated symptoms of prostatism. A further 4 cases had a bladder-neck obstruction without prostatic hyperplasia, causing slight symptoms. The majority had no disorder of micturition until the onset of the infection, and this bears out the accepted opinion that there is little association with prostatic hypertrophy. The source of the infection has not always been easy to determine. There have been no cases arising from gonococcal urethritis; 2 cases were complications of instrumentation for stricture; 1 of an indwelling catheter for a neurogenic bladder, and 1 followed a cysto-urethrogram to investigate a bladder-neck obstruction. One case occurred in a diabetic with a calculus in the prostatic urethra. In all these 5 cases the infection may be presumed to have been urethral in origin, and in all, coliform organisms were isolated with the exception of the one following a cysto-urethrogram, in which cultures were sterile.

Of the other cases there were 6 of staphylococcal and 1 of streptococcal infection which were undoubtedly metastatic in origin. Of the remainder, 11 were coliform infections and 4 unknown, and as there was no known preceding condition such as urinary infection or instrumentation, the probability is that these infections also were blood-borne.

The symptoms in these cases conformed to those generally described. Analysis reveals the following relative incidence: frequency, 16; dysuria, 14; difficulty, 9; retention, 8; discharge of pus, 7; rigors, 6; perineal pain, 6; hæmaturia, 4; rectal pain, 2; low backache, 2; abnormal leakage of urine from perineum, 1; and from the rectum, 1.

Rectal examination was in most cases the main diagnostic factor, but it appears that the commonest error has been to mistake an abscess causing retention of urine for a simple hypertrophy. This occurred in 4 of the 8 cases admitted in this way. In 3 the error was revealed by cystoscopy and rectal examination under an anæsthetic, and in 1 by the discharge of pus after catheterization. The reason would appear to lie in the distortion of the prostate by downward pressure of the full bladder which makes appreciation of the asymmetry difficult, and the fact that the tenderness of the prostate is masked by the greater pain of the retention itself. Fortunately, the true state of affairs is revealed by cystoscopy before prostatectomy is undertaken, or by examination of the prostate again after decompression of the bladder.

A further difficulty encountered in one case was the differentiation from central necrosis of a rapidly growing transitional-celled carcinoma. In this case perineal exposure and biopsy were required as the gross distortion of the urethra made endoscopic manipulations impossible. In reverse, an abscess, especially one which has discharged and resulted in marked fibrosis, may be mistaken for carcinoma. One case in this series had been treated for a urethro-rectal fistula as a case of carcinoma of the prostate before being referred to the genito-urinary department, when further examination showed that the induration was inflammatory in origin and that the fistula had been caused by rupture of an abscess into the urethra and rectum simultaneously.

Acute caseous tuberculosis of the prostate may also require differentiation and one such case has also recently been treated, although not included in this series. Here the history and urine examination provided the clue.

Urine examination is usually but not always helpful in identifying the causative organism. In metastatic abscess pus-cells are always present, but the urine may be sterile if the abscess has no communication with the urethra. Three cases of staphylococcal and one of coliform abscess are included in this series, which had a sterile urine when first seen.

Perineal aspiration is sometimes advocated for diagnostic purposes. It may be helpful if it provides positive confirmation, but may be misleading if negative. It was used only once in this series and did provide a positive diagnosis.

Urethrography has not been used at all for diagnostic purposes as it has been felt that such information as it provides in these cases can equally well be obtained by clinical examination and cystoscopy.

Many methods have been used for the treatment of these cases. Fifteen cases come under the heading of conservative, as spontaneous discharge occurred before or during treatment; in 13 this was into the urethra and in one each, before admission, into the rectum and perineum. In 2, palliative suprapubic cystostomy had been performed elsewhere for relief of retention of urine. In all there was some relief of symptoms and resolution of signs with the natural discharge of the abscess, but the final result was not always satisfactory. Eight cases resolved completely leaving behind no residual signs or symptoms that required further treatment, although 1 case returned after 4 years' normal micturition with a further abscess. The other 7 cases were left with residual symptoms of chronic prostatitis, either in the form of recurrent attacks of infection, or fibrosis in the region of the bladder neck. Five of these cases required further surgery in the form of transurethral resection and urethral dilation to clear out pockets of infection or to relieve fibrous obstruction. Conservative treatment therefore appears to involve a considerable risk of incomplete drainage of the abscess.

Rupture into the urethra by instrumentation was performed 3 times. Twice this was by a catheter introduced for retention and both cases were left with residual symptoms. In the third deliberate rupture against a sound was done, but followed shortly by transurethral resection when a persistent abscess cavity was found and resected; this case was left with no residual symptoms.

Perineal aspiration is not advocated as a method of treatment, but in one case of staphylococcal abscess with retention of urine it proved curative when followed by intensive penicillin therapy. This must, however, be considered exceptional.

Perineal exposure and drainage has not been used for abscess, but was used on one occasion for a very large rapidly growing carcinoma thought to be an abscess.

During recent years it has been considered in this Unit that the primary treatment should be by drainage into the urethra by transurethral resection with the McCarthy resectoscope, as originally suggested by Bell (1938).

Accurate diagnosis can be made by visualizing the posterior urethra which reveals the bulging of the affected lobe or lobes of the prostate and can be confirmed by simultaneous rectal examination. Resection is concentrated on this lobe as for adenoma of the prostate, starting anteriorly as advocated by Nesbit (1943). When the abscess cavity is reached there is a copious discharge of pus, of which a specimen may be taken for culture and sensitivity. The pus is washed away by the irrigating stream and the urethral wall of the granulation-lined cavity widely removed. Any further loculi can be detected by a finger in the rectum and similarly unroofed. No attempt is made to do a complete prostatectomy and the deep wall of the abscess cavity is not transgressed. If necessary, remaining pieces of adenomatous tissue can be removed at a later date after inflammation has subsided.

This procedure fulfils the generally accepted surgical principles of drainage of abscesses through the most adjacent epithelial surface without infringing the protective leucocyte barrier. It has the advantage over other methods of urethral drainage that complete drainage is ensured, and over perineal and suprapubic drainage that no normal tissue-planes are opened up, and that no drains are required. In practice, it has not been difficult to perform by those trained in transurethral surgery.

Nine cases have been treated in this manner. All had required direct admission on account of acute retention or extreme difficulty of micturition. The cases with retention were catheterized before resection was undertaken, and in all appropriate antibiotic cover was provided as a preliminary to operation. This may be an unnecessary precaution, for the theoretical danger of spreading the infection is slight.

Convalescence has invariably been rapid and comfortable for the patient, with immediate relief of all local and constitutional symptoms. The post-operative catheter can be removed after 24 or 48 hours and the patient is fit to return home after some 10 days. Follow-up has shown no residual ill-effects in the nature of recurrent prostatitis or bladder-neck fibrosis.

A typical case history is given.

CASE REPORT

H. G., aged 42 years. Admitted as an emergency with acute retention of urine, complaining of malaise, backache, and rigors for 4 days, with extreme difficulty of micturition and frequency for 2 days. No previous urinary disturbance. On examination he was in great distress, temperature 101°, tense bladder distended to umbilicus. The prostate showed a normal left lobe and a swollen, tender, fluctuant right lobe. Urine showed many pus-cells and culture revealed *Staph. aureus*. He was treated by immediate indwelling catheter and sulphonamide. Three days later cystoscopy showed a normal bladder and a prostatic urethra distorted by a bulging right lobe. Resection of the right lobe liberated quantities of thick pus. The catheter was removed after 48 hours and he was discharged after 14 days. When followed up in out-patients, micturition was completely normal and the prostate was also normal to palpation.

Epididymitis appears to be the only likely complication, as emphasized by Bell (1938) and confirmed in this series. Vasectomy is therefore wise unless the young age of the patient makes the risk of epididymitis worth taking to preserve potential fertility.

Two lessons may be learnt from this survey. Conservative treatment in expectation of spontaneous discharge is fraught with the danger of establishing chronic prostatitis which is subsequently difficult to treat; therefore hospital admission for surgical drainage is indicated more frequently in those cases in which symptoms are not sufficiently severe to make it imperative. Transurethral resection is a satisfactory and safe method of treatment where facilities exist for its performance; it gives the least discomfort and the most rapid convalescence for the patient with minimal late sequelæ. The alternative still appears to be wide perineal drainage.

SUMMARY

1. Twenty-seven cases of prostatic abscess have been reviewed with special reference to the results of treatment.

2. Conservative treatment is often successful in obtaining immediate resolution, but results in chronic prostatitis in a high proportion of cases.

3. Early drainage by transurethral resection is advocated.

I have to thank Mr. A. Wilfrid Adams, Mr. Ashton Miller, and Mr. J. P. Mitchell for allowing me to review these cases, which were under their care.

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PARTIAL HEPATECTOMY FOR PRIMARY LIVER CARCINOMA

BY A. GOUREVITCH, J. W. ORR, AND A. G. W. WHITFIELD

THE extent of the literature on primary carcinoma of the liver suggests that it is a common condition, and indeed it is among the Bantus in Africa, the Javanese, Chinese, Japanese, Indians, and Filipinos. Its frequency among these races seems to be associated with the high incidence of hepatic cirrhosis ; in white-skinned people primary liver carcinoma is extremely rare, but when it does occur it also shows an association with cirrhosis. Ten per cent of sufferers from hæmochromatosis develop primary malignancy in the liver (Stewart, 1931). It may occur at any age from infancy (Castle, 1914 ; Griffith, 1918 ; Pirie, 1932 ; Benson and Penberthy, 1942) to senescence (Hicks, 1929 ; Strong and Pitts, 1930).

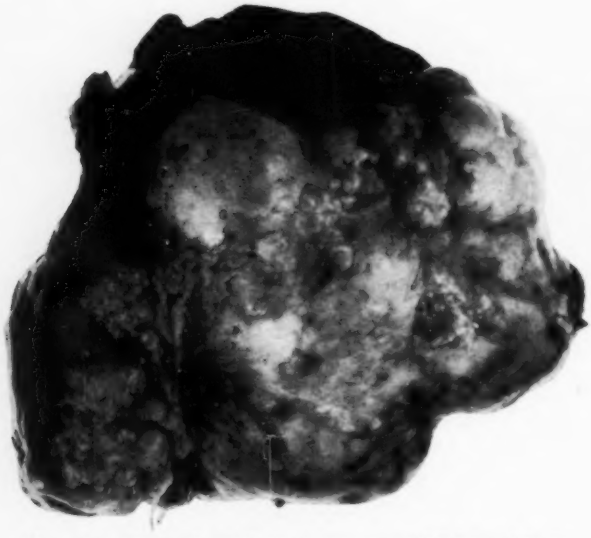

FIG. 725.—External naked-eye appearance of left lobe of liver after removal. (× ⅛.)

FIG. 726.—Naked-eye appearance of cut surface of left lobe of liver consisting almost entirely of a large hepatoma.

Liver-cell carcinomas (hepatomas) are twice as common in this country as tumours arising from the bile-ducts (cholangiomas) (Stewart, 1931). Primary liver cancer tends to spread by venous channels. Involvement of the portal venous system produces metastases in the liver. Involvement of the hepatic veins and inferior vena cava produces secondaries in the lungs, and sometimes tumour thrombi extending to the right side of the heart (Counseller and McIndoe, 1926 ; Fox and Bartels, 1928 ; Willis, 1953). This tendency to the development of metastases in the liver has been advanced as evidence against the theory of multifocal origin. In the recorded cases the patient has usually noticed the mass and complained of some degree of discomfort or pain related to it (Garnett Wright, 1922 ; Fox and Bartels, 1928 ; Abel, 1933 ; Love, 1934–35 ; Wallace, 1941 ; Pickrell and Clay, 1944). Usually the mass is readily felt and often easily seen (Keen, 1899 ; Benson and Penberthy, 1942). Occasionally it ruptures and produces intraperitoneal hæmorrhage (Turner, 1922 ; Strong and Pitts, 1930). The prognosis is extremely poor, but in occasional cases the growth has been successfully excised (Keen, 1899 ; Freeman, 1904 ; Yeomans, 1909 ; Castle, 1914 ; Garnett Wright, 1922 ; Hicks, 1929 ; Le Grand, 1932 ; Pirie, 1932 ; Abel, 1933 ; Love, 1934 ; Goldberg and Wallerstein, 1934 ; Wallace, 1941 ; Benson and Penberthy, 1942 ; Pickrell and Clay, 1944 ; Brunschwig, 1955) and follow-up has shown survival for periods of 16 months (Freeman, 1904), 2½ years (Garnett Wright, 1922), 5 years (Wallace, 1941), and 9 years (Yeomans, 1909, 1915) without recurrence. The purpose of this communication is to record a further case of successful

removal of a hepatoma involving most of the left lobe of the liver, in a male British patient.

CASE REPORT

The patient, a male company director of 46 years, had had no previous illness except for two attacks of bacillary dysentery while serving in the army during the 1939-45

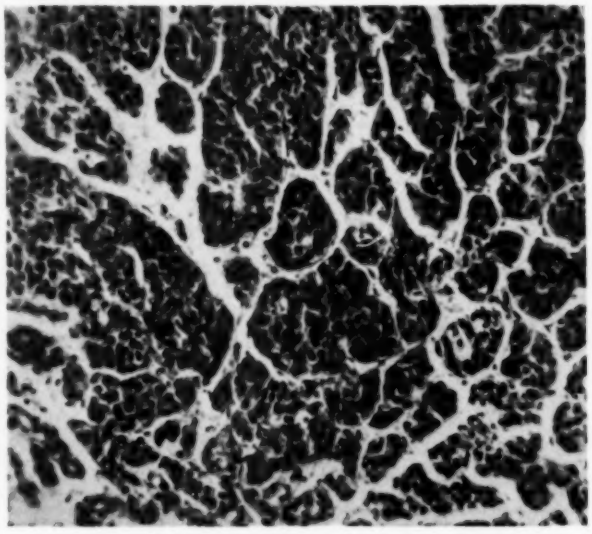

FIG. 727.—Section of tumour to show arrangement of cells in trabeculæ separated by vascular spaces resembling liver sinusoids. H. and E. (× 80.)

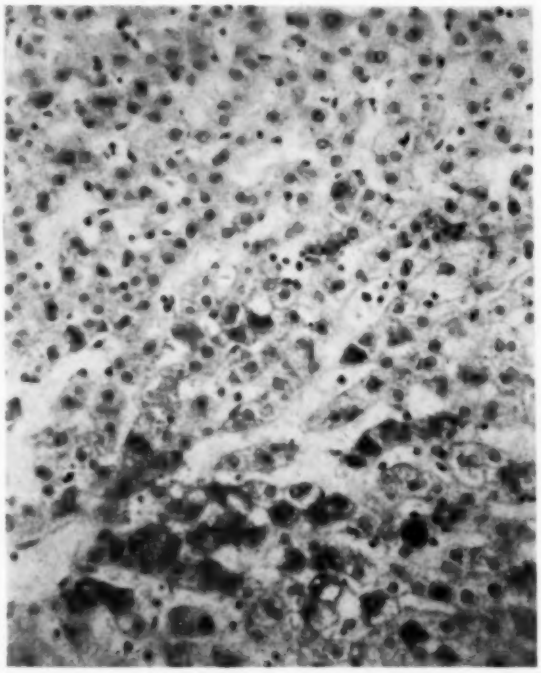

FIG. 729.—Non-neoplastic liver stained for glycogen, which is present in cells in the lower part of the figure. The tumour was devoid of glycogen. Best's carmine stain. (× 150.)

war. He had never had jaundice, and his intake of alcohol had never been excessive. For 4 months he had had discomfort in the upper abdomen. This he had noticed particularly when coughing, when lying on his face at the Turkish baths, and when leaning forward in the bath to turn the tap. He had not felt ill or lost weight and there had been no other symptoms.

ON EXAMINATION.—A large hard irregular liver extending to the umbilicus. Jaundice, ascites, and œdema were absent. The spleen was impalpable and there were no spider nævi or 'liver palms'. No primary neoplasm was evident and the remainder of the physical examination was wholly negative. Except for a high cholesterol (444 mg. per cent) the liver-function tests gave values in the normal range, and the prothrombin was 100 per cent. The Casoni test and blood Wassermann were negative. The sedimentation rate was 4 mm. at one hour (Wintrobe). The stools were negative for occult blood and no pus or pathogens were found in them.

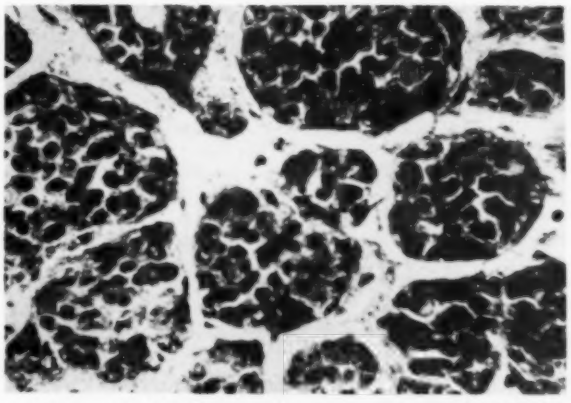

FIG. 728.—Higher magnification to show the delicate endothelial lining of the sinusoids. There is no collagenous stroma. H. and V.G. (× 210.)

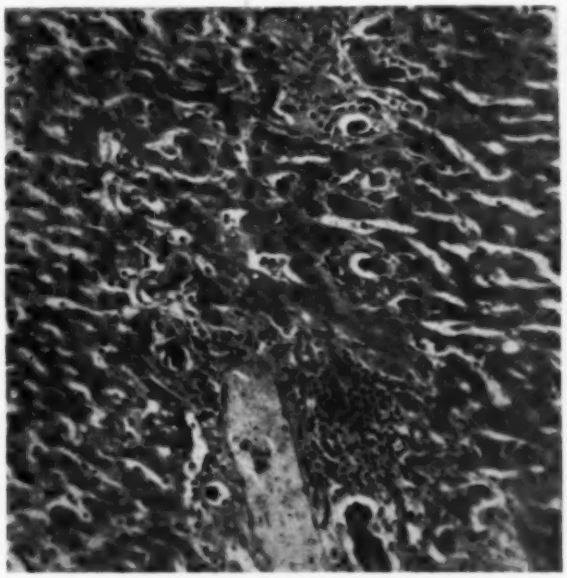

FIG. 730.—Section of part of liver removed with the tumour, showing slight cell infiltration of a portal tract, and outgrowth of a few regenerative bile ducts into the parenchyma. H. and V.G. (× 90.)

The barium enema showed a normal colon and the barium meal revealed a normal stomach and duodenum and no œsophageal varices. The chest radiograph was clear and a complete blood-count showed no abnormality. The urine contained neither albumin nor formed elements. Although it was thought that the liver was the seat of secondary carcinoma, the absence of a detectable primary growth and the patient's apparent good health made us advise a laparotomy.

AT OPERATION.—The right lobe of the liver was normal, but the left lobe was almost completely replaced by a large whitish-yellow hæmorrhagic tumour. Here and there small isolated 'rosettes' of tumour tissue could be seen adjacent to the main mass (*Fig.* 725). The greater omentum was partially adherent to the tumour. Further exploration did not reveal any other tumour or abnormality in the abdomen. This fact, together with the apparently localized and peculiar character of the liver tumour, raised the question of a primary liver cancer. Frozen sections confirmed this suspicion, and the left lobe of the liver was therefore resected. Resection was carried out chiefly by the knife, with diathermy coagulation of vessels. The cut surface of the liver was sutured to the falciform ligament, and the suture line reinforced by the greater omentum. The cut surface of the specimen is shown in *Fig.* 726. The patient made an excellent recovery from the operation and a year later there is no sign of recurrence.

HISTOLOGY.—The tumour shows the characteristic structure of primary liver-cell carcinoma, i.e., polyhedral cells arranged in cords, or trabeculæ separated from one another by capillary channels resembling the liver sinusoids (*Fig.* 727). The trabeculæ are usually solid, occasionally luminated. The stroma is in the main limited to a delicate endothelium which lines the sinusoids and invests the trabeculæ (*Fig.* 728). This characteristic vascular arrangement, the "Kapillarnetz" of Yamagiwa (1911) was also emphasized by Fried (1924) and Bonne (1937), and is insisted upon by Berman (1951) as the distinctive histological feature of liver-cell carcinoma. Collagenous stroma is only found as bands which transect the tumour at wide intervals. The tumour cells are grouped in such a way as to mimic liver tissue, but are never arranged in characteristic lobules. The individual tumour cells resemble liver cells, especially in their nuclei which contain one to three large nucleoli. The tumour cells do not contain glycogen, which can be demonstrated in the non-neoplastic liver tissue of the specimen (*Fig.* 729). Although the latter shows no gross cirrhosis, there is microscopical evidence of some loss of lobular architecture, cellular infiltration of portal systems, and regenerative outgrowth of bile-ducts beyond the confines of the portal systems (*Fig.* 730).

The histology of the tumour has been described in some detail because the view has sometimes been expressed that the diagnosis of primary liver cancer can only be established by necropsy (Willis, 1953).

The absence of glycogen from the tumour cells, while it could be demonstrated readily in the surrounding liver, is a feature which parallels observations (Orr, 1940) on the liver-cell carcinomata experimentally induced in rats by p-dimethylaminoazobenzene (butter yellow). In the small amount of relevant post-mortem material available to us from previous cases glycogen has never been found in the tumour cells, but it may also be absent from, or depleted in, the liver itself. The opportunity of examining surgical material from a well-nourished subject is therefore of value in relating experimental findings to the disease process in man. For similar reasons, attention is drawn to the slight changes in architecture in the non-neoplastic parts of the liver of this patient, because there is reason to think that experimental tumours may arise at a time when the cirrhotic process is still reversible. It seems clear that at no time in the present case were any of the clinical manifestations of cirrhosis in evidence, but we have no means of knowing how far the hepatic damage must advance before it gives rise to the characteristic effects of portal obstruction.

COMMENT

The normal reaction on examining this patient's abdomen was to make a firm diagnosis of secondary carcinoma of the liver and limit treatment to the use of analgesics and opiates, when required, for the relief of suffering. However, the patient's excellent general condition and the absence of any primary neoplasm or secondary growths elsewhere on clinical and radiological examination led us to advise a laparotomy. Even when the abdomen was open the first impression was of massive involvement of the liver by secondary neoplasm. It was only when the exploring hand showed the growth to be limited to the left lobe of the liver, and no primary growth in the abdomen, that the question of partial hepatectomy was considered. The operation proved to be a very much easier matter than was anticipated and the excellent result to date has amply justified its performance.

The case is reported in the hope that it may help to prevent similar cases being dismissed as hopeless and denied a laparotomy.

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A MALIGNANT SOFT-TISSUE PARAGANGLIOMA OF THE LEG

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A MALIGNANT paraganglioma arising in soft tissues is a rare tumour, though a number of cases have been described under a variety of names such as liposarcoma, secondary renal carcinoma, granular myoblastoma, angiosarcoma, etc. The histological appearances of a paraganglioma are, in fact, very typical, and can be recognized quite readily, if the group of tumours to which it belongs is borne in mind. The case here described is very similar to the one published by Randall and Walter (1954), but this type of tumour does not appear to have been widely reported in the British literature, and it seems worth while describing a further case.

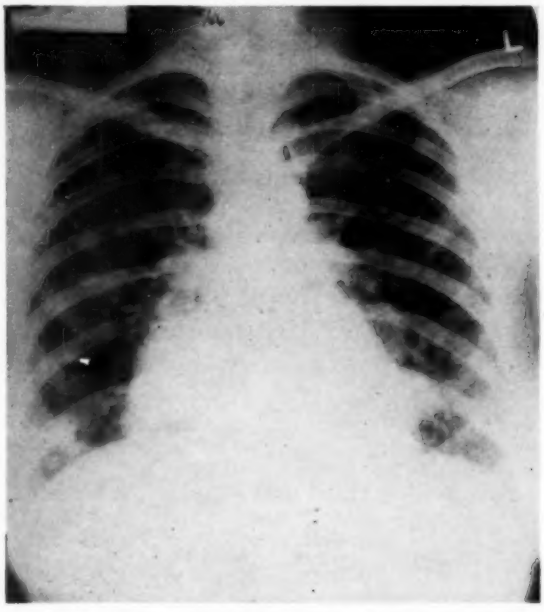

FIG. 731.—Chest radiograph showing secondary deposits.

CASE REPORT

P. L., a housewife aged 22 years, was first seen in May, 1953, complaining of a lump in the upper part of the left calf. The lump had been present for 9 months and was first noticed while she was pregnant; it had slowly increased in size, but caused no disability in walking. There was no history of trauma. There had been some slight pain in the calf, but she had no other symptoms.

ON EXAMINATION.—Radiography of the leg showed a large soft-tissue swelling with no translucency, suggestive of an intramuscular lipoma. Radiography of the chest showed possible early secondary deposits.

AT OPERATION.—The lump was explored through a midline incision splitting the two heads of gastrocnemius. It appeared to be involving the upper part of the soleus muscle, and to the naked eye was fairly well circumscribed. The mass was widely excised without damage to neighbouring nerves.

POST-OPERATIVE PROGRESS.—The patient was seen at intervals following the operation and one year later there was a definite, but slow-growing, local recurrence at the upper end of the calf and radiography of the chest showed numerous rounded secondary deposits (*Fig.* 731). Eighteen months after the operation she remained reasonably well with few symptoms, but in February, 1955, she was re-admitted to hospital with a chest infection. She had been suffering from vomiting and constipation and there was a definite paresis of the right arm. By this time she was pale and emaciated. Her mental condition deteriorated and cerebral secondaries were diagnosed. She went steadily downhill and died in her sleep on April 19, 1955. Permission for autopsy was unfortunately refused.

PATHOLOGY.—The tumour was roughly oval in shape and measured 10 × 6 × 5 cm. The cut surface was a buffy-grey colour mottled with yellow and there were several moderate-sized hæmorrhages (*Fig.* 732).

Microscopic examination showed a framework of vascular fibrous tissue in which were groups of cells separated by fine strands of fibrous tissue or thin-walled blood-channels (*Figs.* 733, 734). The groups of tumour cells varied considerably in size, the smallest consisting of 5 or 6 cells, while others were very much larger. In some places the central cells of the group were missing and this produced a pseudotubular appearance, which is probably an artefact due to the falling out of the central cells during the process of preparing the sections (Christopherson, Foote, and Stewart, 1952). The individual cells were round or polyhedral and measured 20–60 μ in diameter. The cell boundaries were ill defined. The cytoplasm was acidophilic and granular and frequently contained vacuoles. The eccentric nuclei were round and vesicular, though dense pyknotic nuclei were occasionally seen. There were large areas of necrosis and some hæmorrhages. Clumps of tumour cells could be found in small blood-vessels in the fibrous tissue (*Fig.* 735). Mitotic figures were very scanty, but there were occasional giant cell forms with densely staining nuclei. Frozen sections showed occasional droplets of fat in the cell cytoplasm. Sections stained by Laidlaw's silver impregnation technique showed reticulin fibres surrounding the nests of tumour cells but not penetrating between them.

FIG. 732.—The macroscopic appearance of the tumour. (× ½.)

DISCUSSION

It has become evident in recent years that tumours of non-chromaffin paraganglia are more common than was previously thought. First described and best known are those arising in the carotid

body; even these are uncommon. About 320 cases have been described in the literature (Gaffney, 1953) and excellent reviews have been published by Gratiot (1943) and Lecompte (1948). The majority of these tumours are benign; 15–20 per cent are locally invasive, but distant metastases are very uncommon (Gratiot, 1943; Willis, 1948). Similar growths

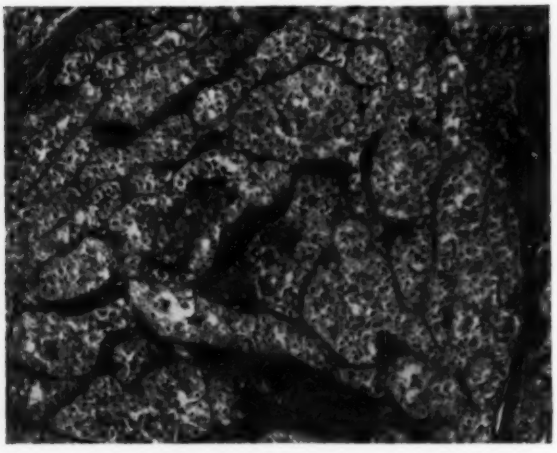

FIG. 733.—A low-power view of the tumour showing the general microscopic appearances. (× 50.)

occasionally arise from the aortic bodies. Six such cases have been published (Davies and Randall, 1954); in 5 of these the tumour was benign, and the sixth produced metastases (Monro, 1950). A third site where this type of tumour is found is the middle ear, in which the growth apparently arises from the glomus jugularis, a structure resembling the carotid body (Guild, 1941). The first description of this type of tumour was published by Rosenwasser (1945) and 56 cases can now be found in the literature (Gaffney, 1953). Some were locally invasive, but none produced a distant metastasis.

In 1951 Smetana and Scott published a description of 14 soft-tissue tumours which they named 'tumours of non-chromaffin paraganglia'. These tumours were histologically similar to carotid-body tumours and it was suggested that they arose from chemoreceptor organs in the soft tissues. The authors quote a personal communication from Dr. C. Johnson who found structures resembling paraganglionic glomera in the tissues around the femoral vessels in Hunter's canal. The tumour described by Randall and Walter (1954) is of the same type as the 14 published by Smetana and Scott, and the one described in this paper belongs to the same group. It is probable that many other similar tumours have been reported under a variety of names without realization of their true origin.

Careful study of the 14 cases of Smetana and Scott, as well as that of Randall and Walter, together with the one described in this paper, reveals a distinctive clinical pattern which may be useful in the diagnosis of this tumour. In the majority of cases the age of onset lies between 21 and 29 years with extremes of 9 and 40 (Smetana and Scott, 1951). The tumour is usually of slow growth, being present for some months before operation. In spite of distant metastases the patient may remain in reasonable health for as long as several years after local excision of the tumour. The commonest site of origin is the thigh, but tumours have also been found in the retroperitoneal region, calf, and neck. The

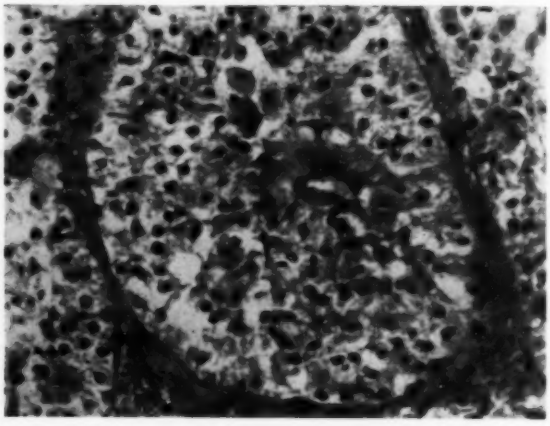

FIG. 734.—A higher power view of the tumour. (× 267.)

growth metastasizes via the blood-stream and secondaries are most commonly found in the lungs. These tumours do not respond to radio-therapy.

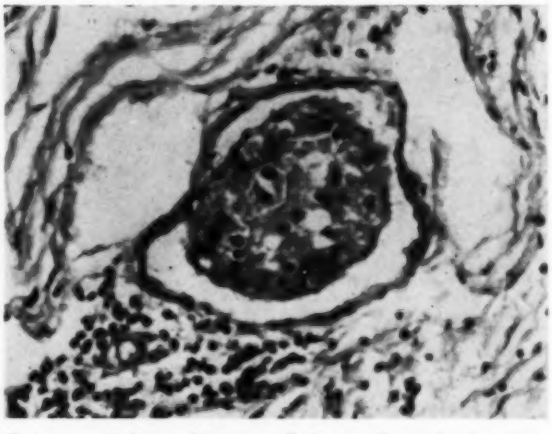

FIG. 735.—A clump of tumour cells in a small vessel. (× 267.)

SUMMARY

A case is described of a soft-tissue tumour of the calf with histological appearances similar to a carotid-body tumour. The growth presumably originated in a chemoreceptor organ in the calf muscles; it produced widespread metastases and resulted in death 2 years and 9 months after the lump was first noticed. Similar previously reported cases are briefly reviewed.

We would like to thank Mr. J. B. Jackson for taking the photographs. The sections were prepared by Mr. E. R. Wood.

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IMMEDIATE POST-OPERATIVE ANURIA IN A HUMAN RENAL HOMOTRANSPLANT

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IN the last decade the treatment of reversible acute renal failure has advanced greatly. In 1946 Lucke estimated the mortality of 'lower nephron nephrosis' (to all intents having the same clinical significance as acute tubular necrosis) at between 80 and 90 per cent. The introduction of careful conservative management has reduced the mortality to the order of 30 per cent (Legrain, 1951; Swann and Merrill, 1953). No such advance has been made in the treatment of chronic renal disease; apart from the improved management of superimposed episodes of acute renal failure, the only advance has been a better understanding of homeostasis by the administration or restriction of various electrolytes, and ensuring an adequate daily urine volume.

With irrecoverable renal damage and insufficient function to maintain life, whether the result of chronic or acute renal pathology, homotransplantation of a kidney is a logical therapy if such a kidney could survive indefinitely and function adequately to maintain life.

In dogs an autotransplanted (i.e., transplanted into another site in the same animal) kidney can maintain life indefinitely, and if the ureter is re-implanted into the bladder normal function results (Dempster, Joekes, and Oeconomos, 1955). Homotransplanted (i.e., from another individual of the same species) kidneys in the dog may function similarly to an autotransplant for a limited period of several days, after which function ceases abruptly (Dempster, 1953, 1954; Hume and Egdahl, 1955; Simonsen, 1953).

A small number of kidney homotransplants have been carried out in man. One case falls into a special category (Murray, Merrill, and Harrison, 1955), being the successful transplantation of a kidney from one identical male twin to the other, the latter having severe bilateral chronic pyelonephritis. That the failure of homotransplanted and homografted tissue is dependent on a gene-transmitted specific immunity is generally accepted. The successful permanent homografting of skin has been used to establish the identity of identical twins (McIndoe and Francheschetti, 1950).

Table I shows the previously reported cases of kidney homotransplants in man. In only 3 of the 22 cases was acute renal failure the indication for attempting homotransplantation (Voronoy, 1936; Landsteiner and Hufnagel, quoted by Hume, Merrill, Miller, and Thorn, 1955; Michon, Hamburger, Oeconomos, Delinotte, Richet, Vaysse, and Antoine, 1953). The first 2 did not function and were removed after 48 hours. The behaviour of the homotransplanted kidney observed by Michon and others was very similar to that described for homotransplantation in dogs; the kidney functioned very soon after transplantation and maintained a good diuresis and excretory capacity, reducing the blood-urea from 100 mg. per 100 ml. in the absence of any other kidney, for 16 days, and then abruptly ceased functioning. A biopsy 18 hours after the arrest of function showed tubular damage and widespread immature plasma-cell infiltration, as had been previously found in dogs.

All the remaining homotransplants were performed in patients with chronic renal disease. Of the 6 who showed any significant function only 2 secreted urine during the first days after operation. One secreted 700 ml. during the first 12 hours, then became anuric and failed to recover function. The other secreted 1700 ml. the first day and 153 ml. the second, was then anuric for 12 days, and recovered function for 23 days, with a maximum output of 1400 ml. on the twenty-third post-operative day; death occurred 10 days after the second arrest of function of the transplant (forty-seventh post-operative day). The other 4 kidneys to function all passed through an anuric phase lasting from 6 to 18 days from the time of operation. The time of certainly effective function after starting a diuresis was respectively at least 10, at least 13, 19, and 144 days. In no instance was the histological picture obtained, either by biopsy or post-mortem examination, typical of that seen in dogs or in the human homotransplant cited above. In 2 cases the kidneys showed widespread thrombosis with infarction. The kidney which functioned very adequately up to 144 days showed relatively normal glomeruli with generalized tubular atrophy and cellular parenchymal infiltration interpreted as 'ischæmic nephrosis'; no thrombosis was present. The fourth kidney was macroscopically essentially normal (19 days after transplantation) except for a small area of infarction.

Apart from that in the boy whose single kidney was removed following traumatic rupture (Michon and others, 1953), no concrete evidence exists of the experimentally found 'homograft' reaction in the few human renal homotransplants so far studied. In one instance (Hume and others, 1955) there is absolute evidence of function, which would

have been adequate to maintain life in the absence of other renal tissue, for a period of 144 days after transplantation. In this instance it seemed, moreover, that the terminal illness was due, not to an alteration of function in the homotransplanted kidney, but rather to left ventricular failure, which led to the decline of its function during the last week of life.

In studies of kidney homotransplantation in dogs (Dempster, 1953, 1955; Simonsen, Buemann,

Table I.—PREVIOUSLY REPORTED CASES

YEAR	REFERENCE	PATIENT'S AGE, SEX, AND BLOOD-GROUP	LESION OF PATIENT'S KIDNEYS	DONOR'S AGE, SEX, AND BLOOD-GROUP	HOW DONOR KIDNEY OBTAINED
1936	Voronoy	26 F. O (1)	Hg poisoning	— M. B (3)	Donor died of encephalitis
1945	Landsteiner and Hufnagel quoted by Hume and others, 1955	Young F. —	Acute renal failure	— — —	Cadaver
1950 1951	Lawler and others	44 F. AB Rh −ve	Bilateral polycystic kidneys	49 F. AB Rh −ve	Donor died after hæmorrhage from œsophageal varices (cirrhosis of liver)
1951 (a and b)	Dubost and others	20 F.	Glomerulonephritis	49 F. —	—
		44 F. A Rh −ve	R. nephrectomy for tuberculosis 23 yr. previously. L. kidney, chronic pyelonephritis with hypertension	42 M. A Rh +ve	Donor executed
1951	Küss and others	44 F. O Rh +ve	1941 L. nephrectomy	34 F. O Rh +ve	Operative removal of hydronephrotic kidney
		22 M. A Rh +ve	Bilateral renal tuberculosis	22 F. A Rh +ve	Operative removal of tuberculous kidney
		48 F. A Rh +ve	Cancer cervix bilateral ureteric obstruction	40 F. A Rh +ve	R. nephrectomy for hypertension
		15 M. —	Bilateral congenital hydronephrosis	40 F. O Rh +ve	R. nephrectomy for hypertension
		22 M. —	Bilateral hydronephrosis	— M. —	Donor executed
1951 (a and b)	Servelle and others	20 F. —	Chronic hypertensive renal disease with single L. kidney	— — —	Donor executed
1953	Michon and others	16 M. B Rh +ve C-Dee, P+MN	Nephrectomy of single R. kidney following traumatic rupture	— — Patient's mother B Rh +ve, CcDee, P+N	Operation
1952 1955 1953	Hume and others Hawn and others	37 M. A Rh +ve	Chronic pyelonephritis with hypertension and uræmia	49 M. A Rh +ve	Kidney obtained at operation for cancer of lower end of ureter
		30 F. A Rh +ve	Chronic pyelonephritis with hypertension and uræmia	48 M. O Rh +ve	Died during operation for hypertension
		43 F. B Rh +ve	Chronic pyelonephritis with hypertension and uræmia	47 F. B Rh +ve	Died during operation for mitral stenosis
		52 F. B Rh +ve	Chronic pyelonephritis and polycystic disease	21 M. B Rh +ve	Died during operation for Fallot's tetralogy
		18 F. O Rh +ve	Chronic glomerulonephritis	2½ F. O Rh +ve	Nephrectomy for subarachnoid-ureteric anastomosis for hydrocephalus
		26 M. O Rh +ve	Chronic pyelonephritis	33 F. O Rh +ve	Nephrectomy for ventricular-ureteric anastomosis for hydrocephalus
		28 F. O Rh +ve	Chronic pyelonephritis	33 F. B Rh +ve	Died during cardiac operation
		57 F. O Rh +ve	Polyarteritis nodosa	33 M. O Rh +ve	Died during cardiac operation
		26 M. A Rh +ve	Chronic glomerulonephritis	37 F. A Rh +ve	Died during cardiac operation
1954	Murray and Holden	— F. —	Chronic glomerulonephritis	Young — —	Cadaver

Gammeltoft, Jensen, and Jørgensen, 1953; Hume and Egdahl, 1955) a consistent pattern of behaviour has been found, with failure of function within 20 days of transplantation. The apparent discrepancy from the results in at least one of the human cases reported by Hume and others would seem to justify further attempts in man.

In the present case no information on the essential problem of homotransplantation has been obtained. The interest lies entirely in the histological picture,

OF KIDNEY HOMOTRANSPLANTS IN MAN

TIME OF ISCHÆMIA	SITE OF TRANSPLANTATION	DEFINITE FUNCTION OF TRANSPLANT. DAYS POST-OP.	REASON FOR CESSATION OF FUNCTION OR REMOVAL OF TRANSPLANT
—	Femorals	0	Patient died 48 hr. post-operatively
—	Brachial artery and cephalic vein	0	Removed after 48 hr. Patient's own kidneys resumed function
—	L. renal fossa after removing patient's own kidney. Ureter into bladder	Impossible to assess owing to mixture of urines in bladder	Still in position but atrophic and function-less 6 mth. later
—	R. iliac vessels. Cutaneous ureterostomy	—	—
1 hr. 50 min.	R. iliac vessels. Cutaneous ureterostomy	11th–13th d. urine volume 180–230 ml. in 24 hr. with urea concentration up to 1 g. per cent	Urine volume fell off during last 3 d. of life. Patient died on 17th d. after transplant
1 hr. 15 min.	Internal iliac artery, external iliac vein. Cutaneous ureterostomy	Maximum 24 hr. urine output 45 ml.	Patient died 35 d. post-operatively
1 hr. 25 min.	ditto	Maximum 24 hr. urine output 72 ml.	Patient alive 5 mth. later. Transplant still in situ
45 min.	ditto	Maximum 24 hr. urine output 80 ml.	Patient died 20th post-operative d.
—	—	—	Operative death
1 hr. 15 min.	ditto	0	Transplant removed 48 hr. later
3 hr. 20 min.	ditto	Slowly increasing urine volume reaching 600 ml. on 19th d.	Patient died 20th d.
55 min.	External iliac vessels. Uretero-ureteric anastomosis	21 d.	Abrupt cessation of function interpreted as homotransplant immune reaction
1 hr. 10 min.	Renal artery anastomosed to splenic artery; renal vein to renal vein; ureter to ureter	Difficult to assess. Did not excrete dye or maintain patient	Died 37 d. post-operatively
2 hr. 30 min.	Thigh	Did not function	Removed 17th d. Found to be infarcted
2 hr. 15 min.	Thigh	Anuria 8½ d. Maximum function 18th–22nd d. Output fair for 53 d. Still excreting some on 70th d.	Patient died 99th d.
3 hr. 20 min.	Thigh	Anuric 11 d. then good diuresis (maximum 2826 ml./24 hr. declining from 29th–65th d.)	Kidney removed 101st d. after virtually no function for 36 d.
55 min.	Thigh	1700 ml. first 24 hr. 153 ml. second 24 hr. Then anuric until 14th d., when diuresis started. Maximum 1400 ml. on 23rd d. Declined steadily from 26th–37th d.; then ceased	Patient died 49th d.
14 min.	Thigh	700 ml. first 12 hr. No subsequent function	Biopsy 19th d. showed marked ischæmia but no evidence of infarction or thrombosis. When removed on 52nd d. there was massive necrosis and infection
3 hr. 20 min.	Thigh	0	Removed 17th d. Totally infarcted
4 hr. 35 min.	Thigh	0	Patient died 38th d. Transplant showed glomerulonephritis (contr donor kidney normal)
3 hr.	Thigh	Anuric for 18 d. Then diuresis with 24 hr. volumes 1500 2000 ml. up to d. 153	Patient died 176 d. after transplantation from left ventricular failure
2½ hr.	External iliacs. Ureter into bladder	Immediate post-operative urine formation. Not possible to assess after this	Kidney said to be viable 15 mth. after transplantation

that of a proximal tubular necrosis, which is interpreted as the result of the temporary total interruption of blood-flow before and during and perhaps after transplantation. This lesion is not regarded as a tissue immunity reaction, and it is argued that it represents recoverable damage.

CASE REPORT

An unmarried girl of 22 was admitted to hospital on May 18, 1955, severely ill with pelvic peritonitis, the consequence of a septic abortion.

On admission to hospital she had been about 16–18 weeks pregnant, and during the previous two months the patient had attempted to procure abortion by several means, including ingestion of unspecified tablets and the repeated use of a sponge soaked in soap solution placed in contact with the cervix uteri. The day before admission to hospital another person had used a Higginson's syringe and injected dettol solution, either intra-vaginal or intra-uterine. Following this the patient developed severe abdominal pain and vomited repeatedly.

On Examination.—An ill febrile girl with slight jaundice. Generalized abdominal tenderness and guarding; uterus enlarged up to umbilicus, with the os slightly dilated and some placental tissue protruding. No bowel sounds were heard.

Investigations: W.B.C.: 15,000 polymorphs. 90 per cent; serum bilirubin 1·3 mg. per cent; plasma Na 126 K 6·9 mEq./l.; high vaginal swab: toxin-producing organism ? *Cl. Welchii* sensitive to penicillin, chloramphenicol, and aureomycin.

Progress: From the time of admission the patient had complete anuria. Penicillin 5,000,000 units 6-hourly, streptomycin 1 g. b.d., 30,000 units *Cl. Welchii* antitoxin I.V., and 100,000 units tetanus antitoxin were administered. Fluid intake was maintained at 1 l. per 24 hours.

May 19, 1955: Blood-urea 325 mg. per cent, Hb 81 per cent.

May 21, 1955: Blood-urea 520 mg. per cent, Hb 49 per cent. Patient afebrile but not so well.

May 22, 1955: Cystoscopy, and bilateral ureteric catheters passed. Bladder empty, some hæmorrhagic areas at base; no urine obtained from renal pelves. Patient became very drowsy, and general condition worse.

May 23, 1955: Transferred to St. Mary's Hospital, Paddington.

On Examination.—A pale, drowsy, and disorientated girl. Blood-pressure 130/30. Venous pressure raised slightly; T. 95·5°. P. 106. R. 30 with deep respiratory excursion. Heart, N.A.D. Lungs, N.A.D. Abdomen generalized tenderness ++. No bowel sounds. Uterus enlarged up to umbilicus. Central nervous system, N.A.D.

Investigations: Blood-urea 575 mg. per cent; plasma Na 136 K 7·9 Cl 94 HCO_3 9·8 mEq./l.

May 24, 1955: Patient's water balance and caloric requirements maintained through indwelling superior venal caval polythene catheter with concentrated glucose solution. 250 ml. urine passed, greenish brown in colour, containing many pigmented casts. In the evening a sudden rise in pulse-rate to 120, and a fall in blood-pressure to systolic values below 80, and unmeasurable diastolic levels occurred, associated with multiple extra-systoles.

May 25, 1955: Urine volume reduced to 100 ml. and the urine was very heavily blood-stained.

May 26, 1955: Urine volume 50 ml., still blood-stained.

May 27, 1955: Total anuria.

The operative death of a 52-year-old man, with a normal pre-operative blood-urea, and the same blood-group as the patient, A Rh+, made the emergency attempt to homotransplant a kidney possible.

At Operation (Professor C. G. Rob and Mr. W. J. Dempster).—End-to-end suture of renal artery of left donor kidney to patient's left superficial femoral artery in thigh, and end-to-end suture of renal vein to superficial femoral vein. Ureter brought out through an independent wound medial to the main skin incision. Donor kidney circulation recommenced 2 hours and 50 minutes after presumed death of donor.

May 28, 1955: The post-operative course was complicated by gastric bleeding and a very profound fall in blood-pressure not corrected by transfusion until intravenous hydrocortisone had been given.

During the 5 days that the patient survived after the kidney homotransplant only small amounts of blood-stained urine, up to 100 ml. per 24 hours, were collected. The patient's own kidneys started secreting a small increasing volume of clear urine, reaching 400 ml. the day before her death. Death occurred following inhalation of vomit after a generalized convulsion.

Pathology.—*Transplanted Kidney* (fixed 6 hours after death).—

Gross appearance: The kidney was enlarged, measuring 14 × 8 × 3·5 cm. and weighing 235 g. The capsule stripped easily exposing a smooth pale brown surface. On cutting the kidney, the cortex, which was of normal thickness but pale and mottled with reddish areas, tended to bulge. The medulla showed numerous hæmorrhages especially at the apices of the pyramids. Petechial hæmorrhages were apparent in the pelvis.

Microscopical appearances: The glomeruli were normal and showed no appreciable differences from those of the contralateral donor kidney (*Fig.* 736). There was no evidence of inflammation or increased cellularity of the tufts. In occasional glomeruli the parietal epithelial cells of Bowman's capsule were cuboidal, but this was to be seen with equal frequency in the control kidney. Also, in both transplanted and contralateral donor kidneys Bowman's spaces were dilated, this dilatation extending into the first part of the proximal tubules.

Necrosis was clearly apparent in the proximal convoluted tubules affecting the majority of the nephrons (*Fig.* 738). Some of the cells had shrunk down to form a pink staining granular rim to the tubular lumen, others were swollen, with pink granular cytoplasm, and were in process of disintegrating and falling into the lumen. Remains of nuclei could still be clearly recognized. Occasional tubules appeared to have ruptured. Some of the epithelial cells of the proximal convoluted tubules showed signs of regeneration, a few with basophilic cytoplasm forming irregular syncytial masses, others showing mitotic figures (*Fig.* 742). The lumina of practically all these tubules contained pink staining granular casts which were P.A.S.-positive.

The loops of Henle showed no necrotic lesions, only an occasional basophilic epithelial cell. Both limbs of the loop contained large numbers of pink granular casts, and some casts composed of desquamated epithelial cells. The distal convoluted tubules and the collecting tubules contained numerous hyaline and cellular-debris casts, but showed no obvious necrosis of their lining epithelial cells.

Nephrons from this kidney, dissected by Dr. Darmady, showed a very severe necrosis of the first part of the proximal convoluted tubules with rupture of the basement membranes (*Fig.* 745). This change also affected the neck, but there was no narrowing comparable to that seen in dog-homotransplanted kidneys (Darmady, Dempster, and Stranack, 1955). Less severe degenerative changes were seen in the lower part of the proximal convoluted tubules, and very occasional focal areas of necrosis in the loops and distal convoluted tubules.

Cysts were present as in the control transplanted donor kidney. The interstitium was moderately œdematous and there was a mild focal infiltration by lymphocytes, histiocytes, some plasma-cells, and occasional

eosinophils. This infiltration was sometimes seen around necrotic tubules which had ruptured, sometimes adjacent to cysts, but there was no consistent relationship between the inflammatory cells and any structural unit in the kidney. The infiltration was a little heavier than in the control non-grafted kidney. Areas of interstitial hæmorrhage were seen, most commonly at the apices of the pyramids.

One papilla had undergone a hæmorrhagic infarction. No vascular cause for this was found.

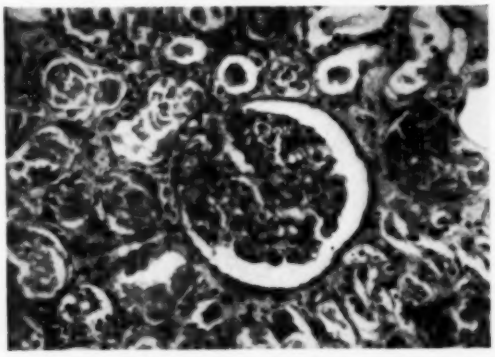

FIG. 736.—Homotransplanted kidney showing normal glomerulus. There is extensive necrosis and desquamation of epithelium of surrounding proximal convoluted tubules. Hæmatoxylin and eosin. (× 96.)

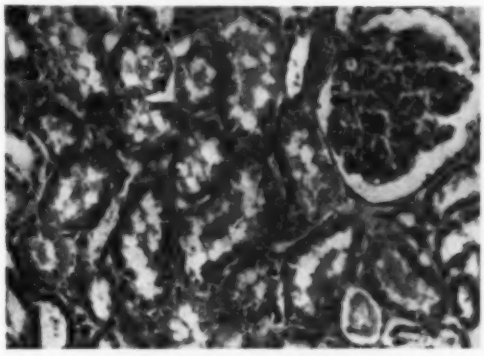

FIG. 737.—Control non-grafted donor kidney. There is early post-mortem change in the lining epithelial cells of the proximal convoluted tubules. Hæmatoxylin and eosin. (× 96.)

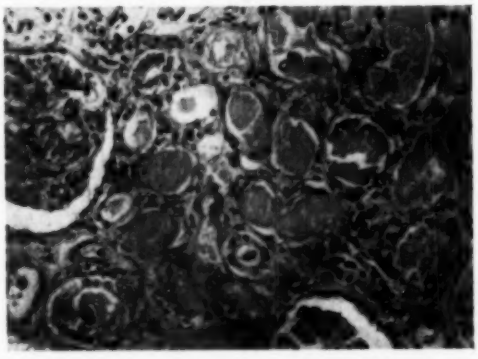

FIG. 738.—Human homotransplanted kidney showing selective necrosis of epithelium of proximal convoluted tubules with cast formation. Hæmatoxylin and eosin. (× 96.)

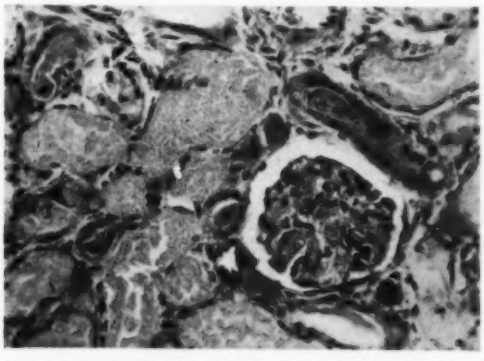

FIG. 739.—Scarff and Keele's photomicrograph of kidney of rabbit after two hours' complete obstruction to renal blood-flow.

There was fibrous intimal thickening of the interlobar and arcuate arteries, but no more than in the non-transplanted control kidney. The interlobular, afferent, and efferent arterioles were normal. None of these vessels was thrombosed, or their lining endothelium swollen. The venæ rectæ and peritubular capillaries were congested, and some of the venules of the medulla showed intravascular accumulations of mononuclear cells. Similar accumulations were seen in the patient's own kidneys.

The anastomosis between the femoral artery and the renal artery was completely patent, there being no thrombosis. The renal artery was normal except for occasional small fibrous intimal plaques similar to those in the contralateral donor kidney. A small non-occluding thrombus, consisting largely of platelets with a few red cells and polymorphonuclears, was present at the site of the anastomosis between renal and femoral veins, and extended for 2 cm. along the renal vein.

The pelvis showed a few submucosal hæmorrhages, but the epithelium was normal. There was patchy loss of the lining transitional epithelium of the ureter and the whole wall was infiltrated with lymphocytes, plasma-cells, macrophages—many containing blood pigment—and a few polymorphonuclears. Submucosal hæmorrhages were again frequent. However, apart from these changes, most of the epithelium and the muscle-layers looked normal.

The renal capsule appeared normal. There was a thick pericapsular layer of fibrin, red cells, and some polymorphonuclears, which had not yet started to organize.

Non-transplanted Contralateral Donor Kidney (Fixed 3½ hours after donor's death).—

Gross appearance: The kidney was of normal size, measuring 11 × 6 × 2·5 cm. and weighing 180 g. Apart from several smooth lined cysts in cortex and to a lesser extent in medulla, no other abnormality was seen.

Microscopical appearance: The glomerular tufts were congested. In occasional glomeruli the parietal epithelial cells of Bowman's capsule were cuboidal. Bowman's spaces were dilated and this dilatation extended into the proximal convoluted tubules, where the lining epithelial cells showed early post-mortem change with loss of their brush borders (*Fig.* 737). The loops of Henle, the distal convoluted tubules, and collecting tubules were normal except for a very occasional pink hyaline cast. Dissection of nephrons showed no abnormality (*Fig.* 746).

Several small cortical and medullary cysts were present. These were lined by flattened epithelium and contained pink staining proteinous material.

There was a patchy infiltration of the interstitial tissue with lymphocytes and occasional plasma-cells. Some of this infiltration was in relation to the cysts.

The interlobular and arcuate arteries showed some fibrous intimal thickening with increase of elastic tissue. The interlobular, afferent, and efferent arterioles were normal. There was congestion of the venæ rectæ. The main renal artery showed occasional small fibrous intimal plaques.

Pelvis, ureter, and renal capsule were normal.

GENERAL POST-MORTEM FINDINGS (30 hours after death).—

External Appearances.—The body was that of a well nourished young woman. Her breasts showed the dark areolar pigmentation of pregnancy. There was a brownish discharge from the vagina. A kidney had been transplanted on to her left upper thigh and was now surrounded by a layer of blood-clot which was lightly adherent to the renal capsule. Large blisters were present over both heels.

Cardiovascular System.—The heart was normal.

Respiratory System.—The lungs were congested and œdematous with patches of early bronchopneumonia most prominent in the lower lobes. Hæmorrhage had occurred into occasional alveoli.

Digestive System and Peritoneum.—There was a pelvic peritonitis with purulent material lying in the pouch of Douglas. Loops of gut in this area were hyperæmic and coated with exudate. The liver showed 'cloudy swelling' of its cells.

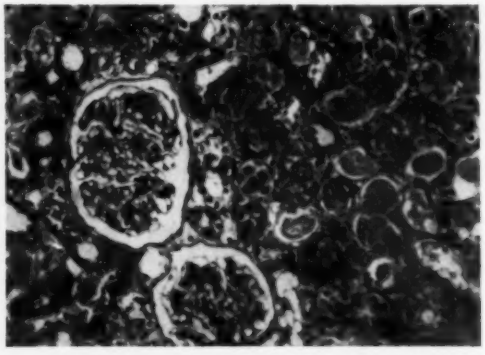

FIG. 740.—Dog-autotransplanted kidney showing similar necrosis of epithelium of proximal convoluted tubules and casts. Hæmatoxylin and eosin. (× 84.)

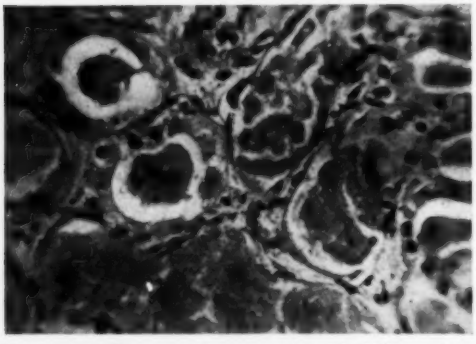

FIG. 742.—Human homotransplanted kidney showing varying stages of degeneration, regeneration, and desquamation of epithelium of proximal convoluted tubules. One lining cell is in mitosis. Hæmatoxylin and eosin. (× 240.)

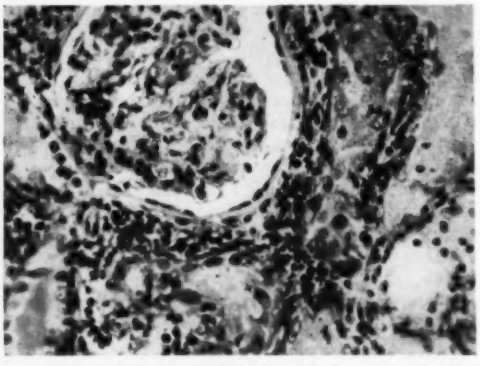

FIG. 744.—Higher magnification of the same dog-homotransplanted kidney as in *Fig.* 743. The proximal tubules are the site of active epithelial regeneration and two mitoses can be seen. Hæmatoxylin and eosin. (× 240.)

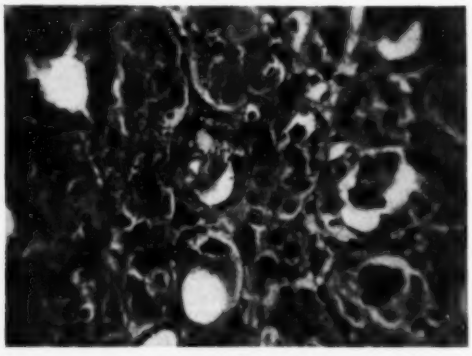

FIG. 741.—Dog-autotransplanted kidney showing similar degeneration and regeneration of epithelium of proximal convoluted tubules. A tubule near the centre of the photomicrograph shows two of its lining cells in mitosis. Hæmatoxylin and eosin. (× 240.)

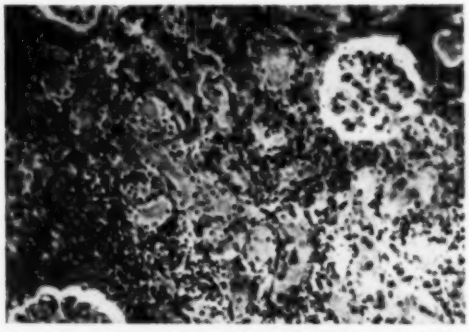

FIG. 743.—Dog-homotransplanted kidney at fifth day. This kidney was anuric from the time of transplantation. Note the pronounced periglomerular immature and mature plasma cell infiltrate. Hæmatoxylin and eosin. (× 84.)

Pancreas, Spleen, Adrenals, Thyroid, and Pituitary were normal.

Genital System.—The uterus was enlarged and soft and its peritoneal surface coated with exudate. There were two perforations at the fundus. The cavity was lined by greenish-black, ragged, friable, necrotic tissue and remnants of a retained placenta. The wall of the uterus was riddled with abscesses and infection had spread to the parametrial tissues, both uterine tubes, and both ovaries. The breasts showed the usual changes of pregnancy.

Nervous System.—The brain was œdematous.

Patient's Own Kidneys.—Showed the typical appearances macroscopically and microscopically of healing acute tubular necrosis with the major damage in the distal convoluted tubules and ascending limbs of Henle's loops. These findings were confirmed by nephron dissection.

DISCUSSION

Experience concerning human homotransplanted kidneys has been set out in the introduction. The discrepancy between the course on the one hand of experimental dog-homotransplanted kidneys and the only documented human kidney transplant for acute renal failure, and on the other of kidneys transplanted

into patients with varying degrees of chronic renal failure, is considered to be an imperative reason for pursuing this problem in suitable clinical situations.

In the case presented the indications for performing a homotransplant were not clear-cut, and in retrospect a renal biopsy taken at the critical time

FIG. 745.—Part of a dissected nephron from the human homotransplanted kidney. Note that the main area of necrosis is in the first part of the proximal tubule. (× 60.)

might have persuaded against it. The presumed diagnosis was an acute tubular necrosis following criminally induced abortion; the patient was anuric for 6 days and then passed 250 ml. of urine in 24 hours, but during the subsequent 48 hours a diminishing quantity of blood-stained urine was passed, and on the 10th day she was again anuric. The interpretation of this late episode of hæmaturia and subsequent anuria was made more difficult by the late stage in pregnancy, the fifth month, when the abortion was induced. The possibility of an extension of an incomplete bilateral cortical necrosis could not be dismissed, although the most likely event was still to be an infarct into one or both kidneys, possibly due to thrombosis following a severe fall in blood-pressure after an intestinal hæmorrhage. Even so the patient's condition suggested she would not stand a much further prolonged anuria. With the severe pelvic peritonitis and persistent ileus neither intestinal nor peritoneal dialysis could be performed. Without any available technique for blood dialysis, i.e., no 'artificial kidney', the possibility of a homotransplanted kidney functioning even for only two weeks or so, as in the boy with the removal of his only kidney, would be of great therapeutic value, giving the patient's own kidneys a further period in which to recover.

Despite every endeavour the interval of ischæmia of the donor kidney could not be kept below 2 hours and 50 minutes, and the transplant did not function during the 5 days that the patient survived after the operation. Three other kidneys which were removed 48 hours after transplantation because they failed to secrete urine are comparable; no histological description of one case is available (Landsteiner and

FIG. 746.—Similar part of a dissected nephron from the contralateral donor kidney for comparison with *Fig.* 745. (× 60.)

Hufnagel); another is described as recalling the lesions in the patient's own kidneys, which were those of mercuric chloride poisoning, but no localization of damage in the tubules is discussed (Voronoy, 1936). The third (Küss, Teinturier, and Milliez (1951), *Case* 5) was interpreted as showing 'a lower nephron nephrosis'.

The principal histological changes in the homotransplanted kidney of the present case were:—

1. Necrosis of the proximal convoluted tubules.
2. Indications of attempts at regeneration of these necrotic tubules.
3. Filling of the lumina of the great majority of the nephrons by granular, anuclear, acidophilic casts resulting from necrotic desquamated epithelial cells.

Such changes have been described previously in experimental animals where temporary complete ischæmia of the kidneys has been produced. Tubular necrosis as a result of temporary occlusion of the main renal artery has been described in the rat (Koletsky and Gustafson, 1947; Koletsky, 1954), rabbit (Litten, 1881; Scarff and Keele, 1943;

Badenoch and Darmady, 1947), and dog (Litten, 1881; Mallory, 1947; Selkurt, 1945; Scheibe, Giraldi, and Vermeulen, 1949).

Similar histological changes may be produced by various toxins and poisons, e.g., uranium (MacNider, 1924), tartaric acid salts (Nicholson, Urquart, and Selby, 1938), mercury (Edwards, 1942), potassium bichromate (Schou, 1944), and potassium chlorate.

Koletsky and Gustafson in 1947 showed that clamping of both renal pedicles in rats produced necrosis of tubules involving principally the proximal convoluted tubules. In general the glomeruli showed no significant morphological change. Regeneration of necrotic tubules was shown by formation of new lining epithelial cells with vesicular nuclei and frequent mitoses.

In a more recent paper Koletsky (1954) produced a similar but unilateral, temporary complete ischæmia, again in rats, by clamping the left renal pedicle. With the opposite intact kidney maintaining life he was able to study the entire natural history of the renal lesion resulting from such ischæmia. He found that a regeneration process followed necrosis of the proximal convoluted tubules and he observed virtually complete repair after periods of interrupted blood-flow lasting up to 4 hours. Repair of the tubular necrosis, however, was followed by progressive renal atrophy, the degree of the latter being roughly proportional to the duration of the ischæmia. The atrophy seemed complete at the end of 3 weeks; the kidney then remained permanently small and unchanged as long as the opposite intact kidney was undisturbed. When the opposite kidney was removed compensatory hypertrophy occurred which was good in the case of the kidney deprived of blood-flow for 2 hours, fair for the 3-hour kidney, and poor or negligible for the organ with 4 hours of complete ischæmia. Thus, as Koletsky says, the renal atrophy of 2-hours ischæmia proved in the rat to be reversible, of 3-hours ischæmia partly reversible, and of 4-hours ischæmia irreversible.

In 1943 Scarff and Keele, working with rabbits, removed one kidney, then clamped the opposite pedicle for varying periods up to 2 hours. Gross degenerative changes were produced in the proximal tubules and numerous granular and hyaline casts were seen in the distal and collecting tubules. The glomeruli were unaffected (*see Fig.* 739). In some cases this degeneration was followed by regeneration and apparent return of the kidney to normal.

Selkurt (1945) and Scheibe and others (1949), working with dogs, similarly found that necrosis of the epithelium of the proximal convoluted tubules was the characteristic lesion following temporary clamping of the renal artery.

In view of the great similarity between these findings in experimental animals subjected to temporary complete renal ischæmia and our findings in a human transplanted kidney, it is suggested that the changes observed in the transplant were solely of ischæmia, the damage to the kidney being due to a temporary period of complete anoxia during which the cells of the proximal convoluted tubules presumably underwent profound chemical alteration. With return of the circulation extensive cellular necrosis occurred. The period of complete anoxia was the 2 hours and 50 minutes which elapsed before the renal blood-flow was re-established in the transplanted kidney.

Can we exclude toxins as a cause of this tubular necrosis? As previously stated, a similar histological appearance can be produced by poisons. Nephron dissection (Oliver, MacDowell, and Tracey, 1951), however, has shown that the necrosis due to poisons, uncomplicated by ischæmia, is confined to the proximal tubules and that the tubular basement membrane remains intact; with necrosis due to ischæmia the damage is predominantly in the proximal tubules, but random disruptive lesions occur elsewhere and the tubular basement membrane is necrotic and ruptured.

In a paper in 1954 Dempster described the changes in anuric dog kidneys following transplantation. He found that in some 5 per cent of auto-transplanted and 16 per cent of homotransplanted kidneys there was anuria from the time of transplantation. More recently Hume and Egdahl (1955) found the incidence much lower in their experimental dog kidney homotransplants, only one out of 66 kidneys showing this early anuria.

Microscopically many of Dempster's auto- and homotransplants with this type of anuria have been found to show varying degrees of proximal convoluted tubular necrosis, with evidence of commencing regeneration of the lining epithelial cells and filling of the lumina of the nephrons with pink casts. *Figs.* 740 and 741 show such a dog-autotransplanted kidney which was removed after a 48-hour post-operative anuria.

From the histological appearance, both in the dog-transplanted kidneys and in our human case, and from Koletsky's animal experiments, one would expect the anuria present from the time of transplantation to be reversible by regeneration of the necrotic tubules if the initial damage were not too severe. This is borne out by the clinical findings of Hume (1952, 1955) and others who recorded 5 cases which regained function after a period of anuria lasting from 9 to 19 days; the case of Servelle, Soulié, Rougeulle, Delahaye, and Touche (1951a, b) also had a diuresis on the sixth day. The pathological lesion must therefore be interpreted as a reversible acute tubular necrosis with mainly proximal tubular damage. Although the time of absolute ischæmia in transplanting a kidney must be of importance, other factors such as persistent intrarenal arterial spasm may be equally responsible for causing this acute tubular necrosis.

In the dog-homotransplanted kidney the picture becomes complicated by the early development of an immature plasma-cell infiltration. In Dempster's series one anuric homotransplanted kidney was not removed until 98 hours after transplantation; this showed proximal tubular damage, but in addition prominent active regeneration with numerous mitoses (*Figs.* 743 and 744). The characteristic peri-glomerular immature plasma-cell infiltration was also present. The latter was strikingly absent in our human case, despite careful search with pyronin stains. In *Case* 3 of Hume and others (1952) the homotransplant was biopsied on the nineteenth day, when the kidney was anuric, and again no mention of any plasma-cell infiltration was made. The kidney examined by Michon and others contained numerous

plasma-cells at the twenty-first day. The time schedule varies from kidney to kidney and so absence of plasma-cells at an early stage is of little significance.

SUMMARY AND CONCLUSIONS

1. A case of an attempted therapeutic homotransplantation of a kidney is described. The donor was a 52-year-old man, who died during an operation. One of his kidneys was transplanted on to the femoral vessels of a young woman with anuria following a septic abortion during the fifth month of pregnancy. Donor and recipient were of the same blood-group.

2. The kidney homotransplant remained anuric or grossly oliguric through the 5 days the patient survived after the operation.

3. Histology and nephron dissection of the homotransplant showed a predominantly proximal tubular necrosis consistent with the change found following a period of temporary complete renal ischæmia. The presence of mitoses and other evidence of regeneration in the tubular epithelium indicated that the lesion was reversible. There was no evidence of a tissue immunological reaction. In the light of these findings the nature of the anuria in the homotransplanted kidney has been discussed.

4. It is emphasized that any knowledge obtained from this unsuccessful attempt at a human homotransplanted kidney is entirely confined to an understanding of the immediate post-operative anuria. No further light has been thrown on the essential problem of homotransplant reaction.

5. The patient's own kidneys showed the typical changes of an acute tubular necrosis with the main brunt falling on the distal convoluted tubules and ascending limbs of Henle's loops, such as is seen with mismatched transfusion, crush syndrome, or following abortion.

6. Previous attempts at human renal homotransplantation recorded in the literature are reviewed.

Acknowledgements.—We wish especially to thank Professor C. Rob and Dr. V. Wyn for their great help and co-operation. Our thanks are also due to Mr. C. T. Ealand for referring the case to us; to Professor Scarff for kindly giving us permission to reproduce *Fig.* 741 from one of his papers; to H.M. Coroner for the Western District of the City of London and Dr. Keith Simpson for their permission to publish the post-mortem findings; to Professor D. M. Pryce and Professor G. W. Pickering for much helpful advice and criticism; and to Mr. A. Beasley for the cutting and staining of numerous sections. The microdissection studies were kindly carried out by Dr. Darmady who was in receipt of an expenses grant from the Medical Research Council.

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A COMPARISON OF TEMPERATURES MEASURED IN THE RECTUM, ŒSOPHAGUS, AND ON THE SURFACE OF THE AORTA DURING HYPOTHERMIA IN MAN

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INTRODUCTION

WHEN body cooling is employed to assist cardiovascular surgery, the temperature of the heart is of considerable importance in determining the possible onset of ventricular fibrillation. The assumption is frequently made that the rectal temperature is a reliable index of the deep body temperature in general, and of the heart temperature in particular. This assumption is clearly unreliable when the aorta is clamped in the thorax with the consequent obliteration of the rectal blood-supply. It is also doubtful whether in any circumstances the measurement of temperature in any one cavity reflects accurately the temperature of distant body sites (Burton and Edholm, 1955; Mead and Bonmarito, 1949). During hypothermia in man, the measurement of intracardiac temperatures is impracticable and indeed dangerous. It was therefore decided to investigate whether the temperatures in the rectum and œsophagus were related to that of the blood in the aorta, and which of these two sites gave the more accurate index of the aortic blood temperature.

METHODS

Observations were made on patients who underwent operations for the resection and grafting of aortic aneurysms, or of thrombosed aorta, the ligation of patent ductus arteriosus, and resection of carcinoma of the œsophagus. Each of these patients was cooled in the following manner. Anæsthesia was induced using intravenous thiopentone and maintained with nitrous oxide and oxygen mixture. Intravenous curarizing agents and pethedine were also given. The whole of the anterior surface of the body was covered with ice-bags and a stream of air directed with fans on to the body surface. The ice-bags were removed about 1½–2 hr. later, the patient being adjudged sufficiently cold. During the operation, the patient could be further cooled or warmed by re-applying ice-bags or bags filled with hot water to the legs and upper thorax.

Temperature Measurement.—Copper constantan thermocouples (28 S.W.G.) were used to indicate the temperature at each site. The thermocouples were totally enclosed in polythene tubes whose external diameter was 3 mm. The reference junctions were immersed in a water-bath, the temperature of which was controlled to within ± 0·01° C. The time lag between the immersion of a thermocouple in a stirred water-bath and its attainment of the water temperature was 20–30 sec. The thermocouples were connected through a two-pole, multiway, all-copper switch to a sensitive galvanometer. Each couple was calibrated before and after every operation. The accuracy of any single reading was better than ± 0·05° C. Later observations, including those on the animals, were made with a Pye three-channel recording thermometer.

The œsophageal thermojunction was placed as near as could be judged at heart level, and the rectal couple inserted 4–8 in. into the rectum. On completion of the operation a third couple was placed on the surface of the aorta and located in this position with a loose catgut suture. It was removed the following day in the manner of a surgical drainage tube. The exact site of this para-aortic couple varied; on some occasions it lay on the aorta under the left ventricle, and on others on the surface of the abdominal aorta. Evidence based on animal experiments will be presented which indicates that the para-aortic temperature is closely related to the temperature of the blood within the aorta.

Blood-flow.—In 3 patients measurements were made of the calf blood-flow, there being reason to suspect that a hyperæmia in the legs could be associated with changes in the temperatures observed. Blood-flows were measured by a venous occlusion plethysmograph, initially filled with water at 34° C. and allowed to cool during the first 2 hr. to about 28° C. Volume changes were recorded electrically (Cooper and Kerslake, 1951).

RESULTS

The Relation between Rectal and Œsophageal Temperature during Cooling.—As the patient cooled, the rectal temperature usually fell more slowly than the œsophageal temperature. In *Fig.* 747 the temperatures recorded during cooling of one patient are shown. It can be seen that there was a difference between rectal and œsophageal temperatures of as much as 2·3° C. at an œsophageal temperature of 31·1° C. The difference between the temperatures at these two sites became less as an œsophageal temperature of 30° C. was approached. *Fig.* 748 shows the corresponding rectal and œsophageal temperature during cooling from 10 cases, plotted against each other.

A line is drawn on which all the points would fall if these two sites were at the same temperature. It is clear that they are not identical over the whole range and that the maximum deviation occurs at œsophageal temperatures between 30 and 33° C. The difference between œsophageal and rectal temperature at this phase ranges from 0·3 to 2·3° C.

Rectal and Œsophageal Temperatures and Calf Blood-flows during the Surgical Procedures.—When the ice-bags were removed, the temperatures continued to fall for a time. The rectal temperature eventually dropped below the œsophageal temperature and remained lower until rewarming was nearing completion. The main phenomenon of interest during this part of the

procedure was the behaviour of the temperature in the œsophagus and rectum as the aortic clamps were released, and the blood-flow to the lower limbs was re-established. *Fig.* 749 shows, on an expanded time scale, the events occurring at this time. The

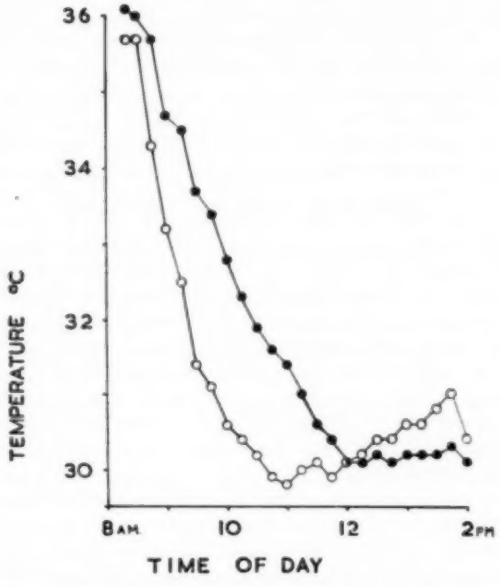

FIG. 747.—Rectal (● — ●) and œsophageal (○ — ○) temperatures plotted against time during cooling of one patient.

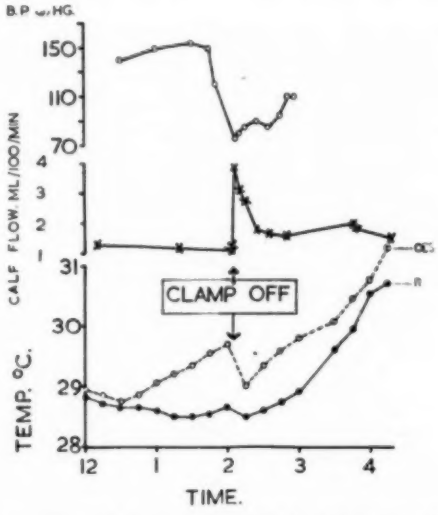

FIG. 749.—Blood-pressure, calf blood-flow, œsophageal, and rectal temperatures on release of the aortic clamps during hypothermia.

removal of the aortic clamps was followed by an abrupt fall in œsophageal temperature of 0·7° C. and a drop in rectal temperature of 0·1° C. There was a large arterial inflow into the calves after the clamps were removed. A precipitate drop of blood-pressure occurred at this time. This arterial inflow settled down to a steady level after about half an hour. Initially, after the clamps were removed, there was no appreciable venous return from the calf, since during this time the vascular bed was refilling. Calculations based on calf volume changes in 4 cases indicate that between 0·5–1·5 l. of blood might be required to refill the vascular bed of the whole of both legs before the full venous return was established.

Temperature during Rewarming.—Patients were placed between blankets in bed on their return to the ward. In most cases no external heat was applied, but in some an electric blanket supplied sufficient heat to prevent heat losses from the patient (i.e., the heater fulfilled the function of more blankets). Before leaving the operating theatre, each

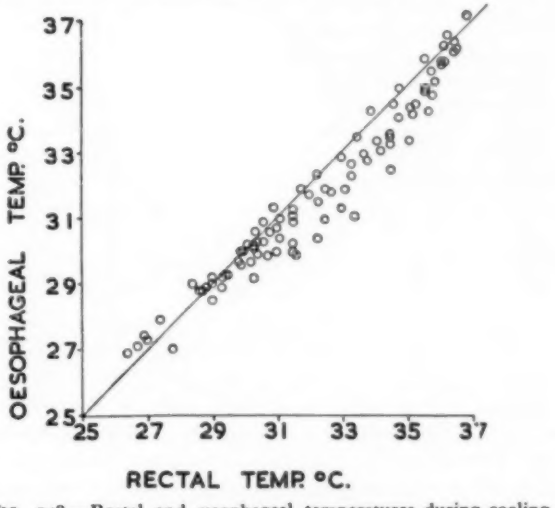

FIG. 748.—Rectal and œsophageal temperatures during cooling (10 cases) plotted against each other. The line is drawn on which points would lie if the two temperatures were identical.

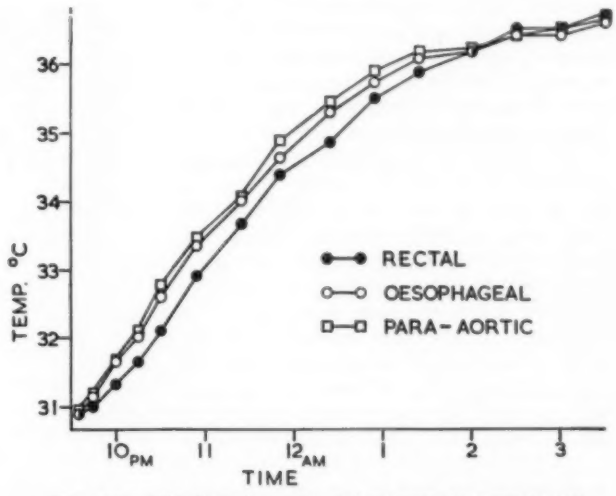

FIG. 750.—Œsophageal, rectal, and para-aortic temperatures plotted against time during rewarming of one patient.

patient received prostigmine intravenously. The rate of rewarming varied over the first four hours between 1 and 2° C. per hour.

Temperatures were measured in the œsophagus, the rectum, and alongside the aorta. *Fig.* 750 shows these temperatures during the rewarming of a patient who had undergone resection of a carcinoma of the œsophagus. In this case the para-aortic thermocouple lay on the arch of the aorta, beneath the left ventricle. The temperature of this latter thermojunction must have been very close to, if not identical with, the temperature of the blood leaving the heart. Œsophageal and para-aortic temperatures followed

each other closely, the maximum deviation of 0·25° C. (0·45° F.) occurring at 11.50 p.m. Rectal temperature rose more slowly and lagged behind the para-aortic temperature by as much as 0·65° C. (1·17° F.). Eventually the rectal temperature exceeded that measured in the other sites. This latter occurrence was frequently observed.

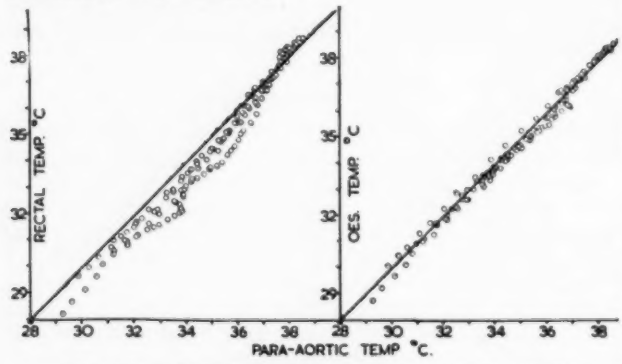

FIG. 751.—Œsophageal and rectal temperatures plotted against para-aortic temperature. Rewarming observations from 10 patients. Each line drawn is that on which all the points would lie if there was no difference of temperature between the two sites of measurement.

The rewarming temperature observations from 10 patients are plotted in *Fig.* 751. Rectal and œsophageal temperatures are plotted separately against the para-aortic temperatures. In each section of the graph a line is drawn on which all the points would lie if there were no disparity between the temperatures. A statistical analysis confirmed what appears evident on inspection of the graph, namely, that the œsophageal temperature was a better index of para-aortic temperature than the rectal.

It might be argued that the temperature of a thermocouple lying on the surface of the aorta could differ from the temperature of the blood within the vessel when heat is being conducted away from the core of the body by ice-bags placed on the body surface. This difference would represent a time lag introduced by the thermal insulation of the aortic wall. Experiments on 2 dogs and 1 pig have been conducted to test the relationship between the para-aortic and intra-aortic temperatures. *Fig.* 752 shows the temperature records made on a dog. The animal was anæsthetized with nitramuscular nembutal and later was given small intravenous doses of thiopentone. The trachea was intubated and respiration maintained with a pump. The animal breathed oxygen, and a CO_2 absorber was included in the circuit. A thermocouple was inserted into the arch of the aorta close to the aortic valves, via a thoracotomy incision, and a second couple was sutured to the outer surface of the aortic arch. The wound was then closed. Cooling was performed by covering the anterior surface of the body with ice-bags and filling the tank on which the dog lay with cold water. The animal was rewarmed by filling the tank with warm water and covering the rest of the body surface with rubber bags containing warm water. In *Fig.* 752 the intra-aortic and para-aortic temperatures taken at quarter-hourly intervals are plotted against each other. The line drawn on the graph is one on which all the points would lie if there were no differences between the intra-aortic and para-aortic temperatures. The difference between these two temperatures during the whole experiment was very small, of the order of 0·1° C., except for two points, which differ by 0·14° C. and 0·13° C. Almost identical results were obtained with the other 2 animals.

GENERAL OBSERVATIONS

Patients became conscious at œsophageal temperatures between 29–34° C. One patient was talking rationally at an œsophageal temperature of 29–30° C. Some details of events at these lower body temperatures were remembered in the later post-operative period when the body temperature had become normal. No marked shivering was observed during the rewarming period, though a certain amount of fibrillary twitching was seen, especially in the pectoral muscles. The limb muscles were usually in a state of severe rigidity up to œsophageal temperatures of 34° C. and considerable restlessness was observed. Almost all patients complained bitterly of cold feet! A few skin temperature measurements made during rewarming indicated that the skin over the trunk was the first to warm up and later the skin temperature in the extremities increased. This increase proceeded peripherally, with abrupt rewarming of wide bands of skin rather than a steady rise over the length of the limb. It was important to realize that the blood-pressure during rewarming was influenced by the rate of rise of body temperature. If this increase exceeded about 1·5° C./hr., a marked

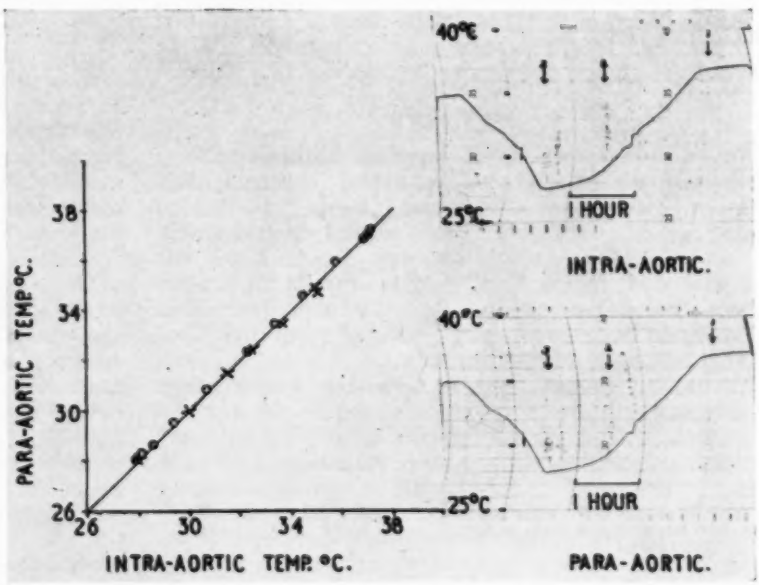

FIG. 752.—Records of intra-aortic and para-aortic temperatures of a dog during cooling and rewarming. Charts read from right to left. The temperature scale on the upper chart reads 0·2° C. low, that on the lower chart being correct. 1, Ice on; 2, Ice off; 3, Rewarming started. Graph: the two temperatures measured at ½-hr. intervals plotted against each other.

fall of blood-pressure occurred which could be restored by removing the blankets from the legs.

DISCUSSION

The particular danger attendant on the use of hypothermia is the sudden and unpredictable occurrence of ventricular fibrillation when the temperature of the body core falls below 27° C. The heart, therefore, is the organ whose temperature must be known accurately during the cooling process. A temperature-measuring device located in the œsophagus at the level of the heart would, by reason of its close proximity, seem to provide a more useful index of heart temperature than would a similar device introduced into the opposite end of the alimentary tract. A simply constructed thermocouple presents no difficulty in its insertion into the œsophagus. Cranston, Gerbrandy, and Snell (1954) have drawn attention to differences between rectal and œsophageal temperatures during experiments which did not involve hypothermia; they state that the rectal temperature is considered a poor guide to the temperature elsewhere in the body.

Under the conditions existing during surface cooling in the pig and the dog, the intra-aortic blood-temperature was reliably indicated by a thermocouple on the outer surface of the aorta (the para-aortic temperature). The evidence presented in this paper leads to the conclusion that œsophageal temperature measured at heart level was a more reliable index of para-aortic temperature than was the rectal temperature. The depth of insertion of the rectal thermocouple between 4–8 in. did not materially affect this conclusion. Making what seems to be a reasonable assumption, namely that the heart temperature must lie close to the temperature of the blood in the aorta, the only tenable conclusion is that the œsophageal temperature is a better index of heart temperature than is the rectal.

Under two circumstances there can be no index of heart temperature. First, when the chest is open and the heart is exposed, for the inside of the organ will be at blood temperature, whereas the external surface will approximate to the theatre air temperature, and a varying temperature gradient must occur across the muscle; the effect of such gradients on the conduction of the excitation wave through the ventricles has yet to be studied. Second, if a large rapid intravenous infusion of fluid at a temperature differing from that of the blood is given, there will be an initial gradient of temperature between the right and left sides of the heart. There is a time lag before such acute changes in heart temperature are reflected in the œsophagus or the rectum, and in our experience the œsophageal temperature gives a more rapid indication of these changes than does the rectal. Even so, the indication given cannot be treated as quantitatively reliable.

It was interesting to note the large hyperæmia occurring in the legs in the hypothermic patient after prolonged ischæmia. The stream of cold blood returning from them should be borne in mind for, if the heart temperature is already about the critical level, the extra cooling may be enough to precipitate cardiac arrhythmias.

The absence of shivering during rewarming was in marked contrast to the events reported as occurring during the cooling of a conscious subject (Burton and Edholm, 1955). Although patients were conscious during this period, residual anæsthetics and analgesics in the body may have been instrumental in the suppression of shivering. A considerable metabolic heat production occurred, for the temperature rose between 1–2° C. per hour for several hours. Although no method is available for the computation of the mean body temperature changes, it is probable that the heat production for a 60 kg. patient was between 40–100 kg.cal./hr. Such metabolic activity was apparent in some patients at œsophageal temperatures of 29° C. and at limb muscle temperatures which were considerably lower. This is of interest in view of the evidence adduced by Wynne (1954) for a breakdown in the mechanism of glucose metabolism at this temperature. The source of energy for such auto-rewarming has yet to be elucidated by further research. Similarly, the significance of the jerky progression of increasing skin temperatures down the limbs is not at present evident.

SUMMARY

1. Measurements of œsophageal and rectal temperatures have been made on patients undergoing hypothermia by surface cooling. Para-aortic temperature was also measured during rewarming.

2. The œsophageal temperature was a more reliable index of para-aortic temperature than was the rectal.

3. Animal studies confirmed the supposition that the para-aortic temperature was a good measure of the temperature of the blood within the aorta.

4. It is concluded that during the type of operation reported the œsophagus was the best site from which to obtain an index of heart temperature.

5. The implications of the temperature changes occurring during cooling and rewarming are discussed.

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PERLMANN'S TUMOUR OF THE KIDNEY

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PYELOGRAPHY during the investigation of his recurrent abacterial pyuria revealed an abnormality in the left kidney of an otherwise healthy 46-year-old ship's officer, who then underwent nephrectomy. He was well 3 years afterwards, though still alleging occasional bouts of pyuria, and in perfect health 6 years afterwards. At operation the surgeon first removed a multilocular cyst and then the kidney itself, since the cyst, when incised in the theatre, showed yellow areas in its walls which reminded him of hypernephroma.

Macroscopic Description.—

1. *Left Kidney.*—No gross abnormality is seen apart from a cavity, containing surgical sutures, situated below and behind the pelvis, corresponding

FIG. 753.—The bisected specimen (photographed 3 years after removal). (× ⅔.)

in shape to the multilocular cyst, and showing no connexion with hilar structures.

2. *Multilocular Cyst.*—This spherical specimen (*Fig.* 753), already incised by the surgeon, measures 4·2 cm. in diameter and shows a mainly smooth, cream-coloured outer surface with a cap of normal-looking brown renal tissue (up to 3 mm. thick) stretched over rather more than one quadrant. Both external and interlocular walls appear to be of similar structure and variable thickness (average 3 mm., though in places reaching 10 mm.), consisting of stratified layers of dense rubbery pearly-white tissue with areas of soft golden-yellow material, often in lenticular shapes, sandwiched between. The main loculi are empty, the largest (2 cm. diameter) being lined by the pearly tissue, which is thinned and bulging in a few places where the yellow material projects slightly into the cavity. One of the smaller loculi appears to be lined wholly by the yellow material, which also fills cystic spaces of pin-head and slightly larger size within the interlocular walls. In these positions it is of paste-like consistency, and gives a positive reaction with fat stains and a strong Liebermann reaction for cholesterol.

Microscopic Description.—

1. *Left Kidney.*—No gross abnormality is seen. A few small wedges of fibrous tissue with round cells occasionally spread inwards from the capsule, and similar areas surround glomeruli in the immediate neighbourhood of the excised cyst, where there is also a zone of operative hæmorrhage.

2. *Multilocular Cyst.*—The cap of kidney substance shows no gross abnormality. The periphery of the multilocular cyst comprises a rind of renal tissue showing compression changes, including marked fibrous endarteritis and tubular distension (*Fig.* 754). There is no sharp distinction between the fibrous tissue of this renal rind and that

FIG. 754.—Rind of fibrosed renal tissue overlies cyst wall containing fibrous tissue, clear cells, and prominent vessels. H. and E. (× 20.)

of the cyst wall beneath it (*Fig.* 754), while surviving cubical epithelial tubules can be found relatively deeply in the cyst wall.

Some cysts contain eosinophil exudate, occasionally with clumps of red cells or desquamated lining cells, like the casts in the tubules of the rind; some are empty, or they may contain fatty debris with large notched cholesterol crystals, sometimes mixed with clumps of red cells and desquamated lining cells. Their lining is of ragged, foamy, cobweb-like cells, frequently shed and usually ill defined. The cells tend to show bent and hollowed contours, which result in their enclosing small spaces and channels (*Fig.* 755). Racquet and withered-leaf shapes occur; hyaline ribbons with almost invisible cell boundaries and ghost nuclei can be found (often partly stripped) as in polycystic and other renal disease; while scanty yet recognizable rows of columnar epithelium with small dense nuclei (as shown in *Fig.* 755) confirm that these linings are indeed epithelial in nature, though masked by the changes of fatty degeneration.

Outside this epithelium there often occurs a zone of similar cells, sometimes foamy, sometimes clear, such as is seen circling the portion of cyst shown in *Fig.* 754, where it appears no different from the adjacent clear-cell area deep in the wall. Within such zones occur many capillary channels which may be bounded simply by filaments of the cobweb cytoplasm of the cells or supported by a network of fine collagen strands. These strands sometimes thicken and unite into short slabs, or parallel

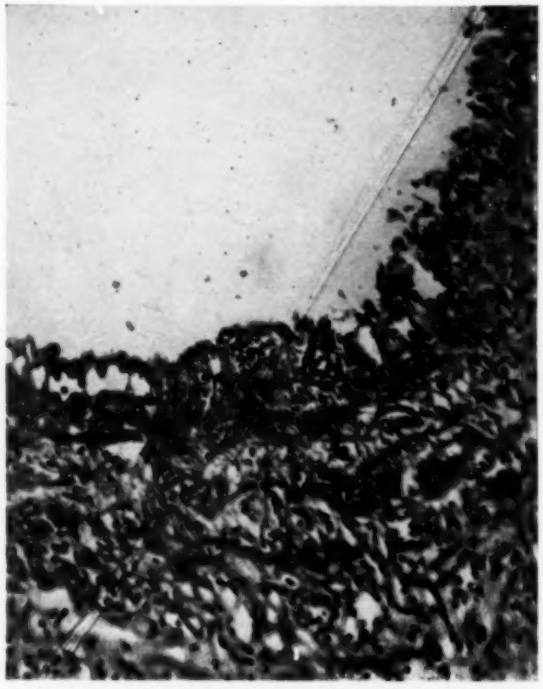

FIG. 755.—Space-enclosing growth habit of an active-looking part of a cyst lining. The spaces towards the centre of the row contain red blood-cells. The epithelial cells show small round dark nuclei. Iron H. and Van G. (× 152.)

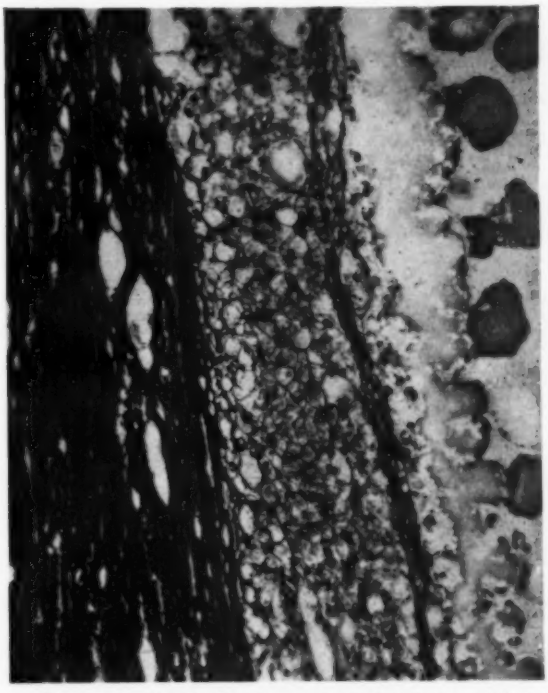

FIG. 756.—Band of collagen separating inner epithelial row from outer lining of clear cells and channels. (Albuminous cyst contents shrunk into cumuli by the silver.) Laidlaw. (× 88.)

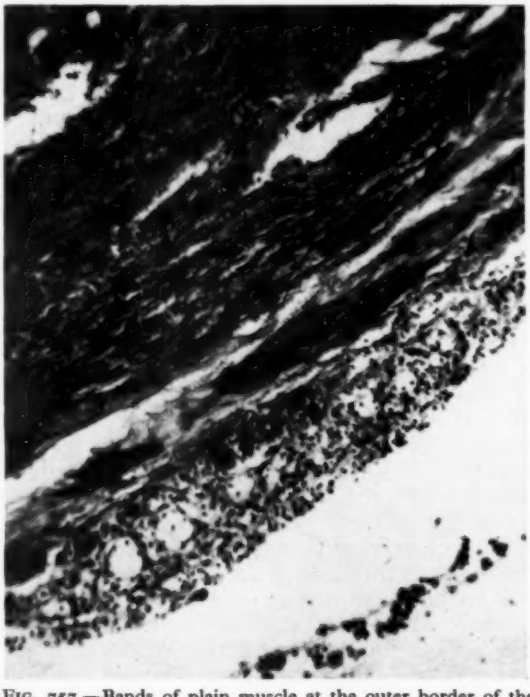

FIG. 757.—Bands of plain muscle at the outer border of the lace-lining of a cyst. Iron H. and Van G. (× 100.)

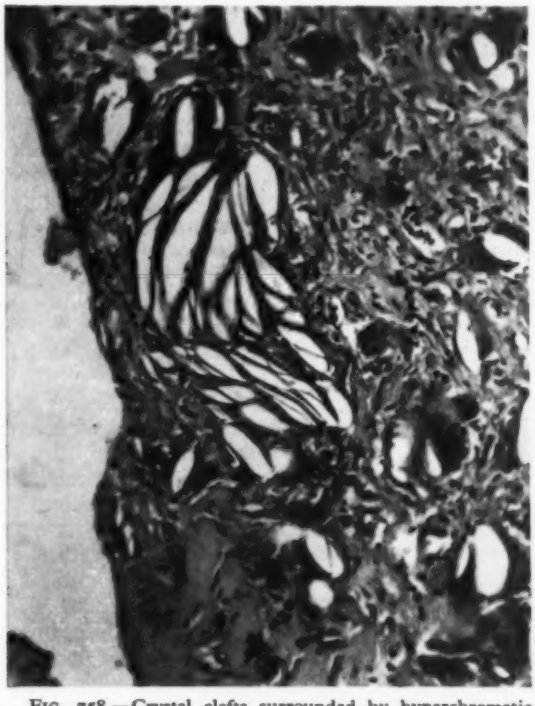

FIG. 758.—Crystal clefts surrounded by hyperchromatic syncytium in an atheroma-like area at the edge of a large cyst. H. and E. (× 128.)

shelf-like formations, or even form a crude basement layer for the innermost columnar row, as in *Fig.* 756. It is these ' lace-linings ' to the cysts which are the striking feature of Perlmann's tumour, differentiating it microscopically from all other renal conditions, and various types of them are shown in *Figs.* 754, 755, 756, and 757. Very rarely, as shown in *Fig.* 757,

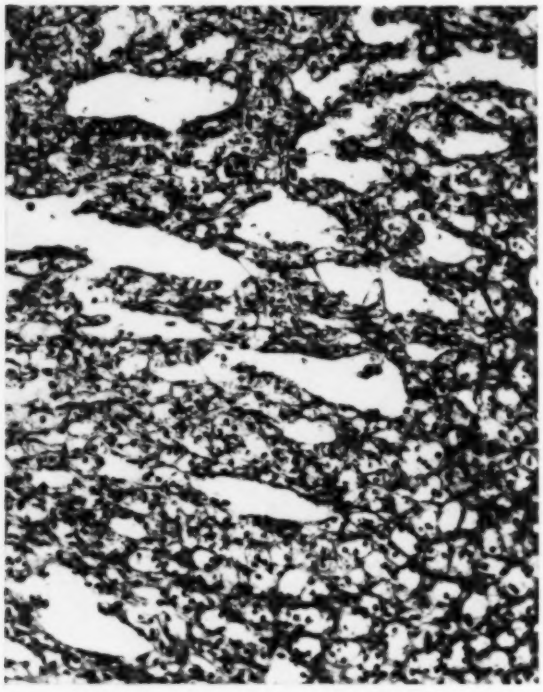

FIG. 759.—Formation of parallel channels in a clear-cell area. H. and E. (× 160.)

concentric lamellæ of plain muscle can also be identified (using Van Gieson) in or near the linings. In places the cysts are lined directly, without any intervening lace network, by atheroma-like hyaline or calcifying fibrous tissue containing fat, in which may be seen (*Fig.* 758) cholesterol crystals surrounded by foreign-body syncytium. Collagen, often more or less degenerate, composes the bulk of the intercystic walls throughout the specimen. In *Fig.* 759 channel-formation by the clear cells has taken the form of parallel sinuses, and in this area the cells resemble unorganized capillary endothelium and even suggest glomerular shapes. Quite large blood-vessels are prominent, but not excessively so, throughout the specimen.

The fatty changes seem to be the chance result of local ischæmia and do not bear any constant relation to the various arrangements of clear-cell strata : in *Fig.* 760, for instance, the Scharlach has principally darkened two zones among the clear cells (that abutting on the renal rind and that lining the cyst) leaving others untouched. Extravasation of red blood-cells into the walls, though it occurs, is not common. Mitotic figures are not seen. Eight blocks were cut before a collection of lymphocytes was found. Stains for elastic and free iron gave negative results. Glycogen staining failed, but was not carried out for over a year after the tissue was fixed.

DISCUSSION

The specimen shows a neoplasia of cyst-forming cells coupled with the processes of fibrosis and fatty degeneration, the latter being common both in renal epithelium and in vascular endothelium. A case can be made out either for the renal or the vascular origin of the clear cells, a quandary reflected in the alternative

FIG. 760.—Intense fatty degeneration in two zones : (*a*) that of a cyst-lining, and (*b*) that abutting on the renal rind. Most of the staining between is of clear-cell nuclei by hæmatoxylin. H. and Scharlach R, frozen. (× 20.)

diagnoses of cystadenoma or angioma offered by most of the pathologists who have kindly helped. Were it not for the neoplastic proliferation of the clear cells, a diagnosis of hamartoma would be reasonable : such tumours, occurring in the kidney with or without generalized tuberosclerosis and sometimes progressing to malignancy, were described in detail by Feriz (1930), who did not, however, figure anything like the architecture of the present specimen. Malignancy cannot be considered among the possibilities in this case, owing to the benign appearance of the clear cells, quite distinct in form and arrangement from those of renal carcinoma.

It is clear from his article that this is the tumour described by Perlmann (1928), and I have been unable to find record of any other. Perlmann's case was that of a woman of 55 with no other symptoms than those mechanically caused by a lump in the left loin. Pyelography proved its renal nature in spite of the absence of hæmaturia. The patient was regarded as cured by nephrectomy. The specimen consisted of a multilocular cyst 11 cm. in diameter, on which were perched the surviving upper pole and pelvis of the kidney. The bright yellow and blue-grey walls of varying thickness (depicted in a colour sketch) and the thinning of the renal substance into a capsule round the tumour resemble the present specimen. Microscopically the ' vesicular cell ' cyst-linings appear identical with those described here, the inner row of columnar foam cells with dot nuclei being well shown (Perlmann, 1928, page 528, Fig. 3), though not described in Perlmann's text.

Perlmann in fact described his tumour as a lymphangioma, but if comparison is made with typical cystic lymphangiomata, such as those described by Goetsch (1938), there appears little justification for this diagnosis, because of the absence of organization of the tissue into endothelial-lined spaces. If the neoplastic cell is considered endothelial, since it is growing in masses the unpopular term ' endothelioma ' would fit better than ' angioma '. A true lymphangiectatic condition at the renal hilum reported by Henthorne (1938) throws doubt, by the orthodoxy of its structure, on the lymphatic affinities of Perlmann's, as of many other peculiar renal tumours, down to the more recent ' lymphangiomata ' of the kidney (e.g., Wynn-Williams and Morgan, 1950). The recent demonstration (Sundarasivarao, 1953) of the adenomatous nature of epididymal lymphangiomata has relevance in this connexion.

Of other possible mesenchymal tumours which might be associated with renal cysts, lipoma and xanthoma can be excluded because of the peculiar ' lace-lining ' structure, although it is true that some isolated clear-cell areas (particularly in thick sections, such as *Fig.* 760) might pass for a rather cellular lipoma, and that the patient had a raised blood cholesterol (350 mg. per cent) when tested 6 months after nephrectomy.

The diagnosis of cystadenoma was picked on as least objectionable for a short published description of the present tumour from the surgeon's point of view (Power, 1955 [in which my pathological description is described as being in the press]). But this term, especially since the papers of Newcomb (1936) and Trinkle (1936), has been so closely associated with one definite series of enlarging renal tumours, only initially cystic, that its use for the Perlmann tumour would be misleading. It seems to me that some kind of double aetiology must be claimed, namely, the supervention of benign epithelial neoplasia (i.e., adenoma) on some congenital defect. In accord with their nature, hamartomata undergoing such change become sarcomata (Feriz, 1930). Perlmann's tumour, on the other hand, shows such similarity to some examples of congenital cystic disease that it might fairly be described as an area of localized polycystic disease which has undergone clear-cell (adenomatous) transformation. This I believe to be the diagnosis. I was assisted to it especially by studying a particular specimen of localized polycystic disease in the Royal College of Surgeons, Edinburgh.

SUMMARY

1. A second case of the tumour described by Perlmann (1928) is described.

2. It is suggested that Perlmann's classification of the tumour as a lymphangioma was incorrect, and that the lesion is produced by the supervention of clear-cell transformation upon localized polycystic disease.

I should like to acknowledge the courtesy of the curator of the Royal College of Surgeons Museum, Edinburgh, Mr. J. N. J. Hartley, in allowing me to study a specimen there, and I should also like to express my indebtedness to Professor Dorothy Russell for her kind help.

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OSTEOCHONDRITIS OF SESAMOID BONES

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ALTHOUGH the clinical manifestations and radiographic findings of osteochondritis of epiphyses are familiar to most, osteochondritis of sesamoid bones is extremely rare.

I have found two contributions written in the English language, one in the Swedish, and another in the British literature. In the first, Renander (1924) has analysed 2 cases of osteochondritis of the medial sesamoid bone of the first metatarsal, and in the second, Johnstone (1942) has reported a case of osteochondritis of the sesamoid bone of the second metatarsal.

I have decided to report this case, not because it is a disabling condition, but because its rarity makes it interesting. As far as I know osteochondritis of the lateral sesamoid bone of the first metatarsal has not previously been described.

CASE REPORT

A girl of 14 attended the Orthopædic Clinic with a history of having had pain in her left foot for about a year. There was no history of any injury to the left foot and apart from the symptoms relating to this, she had always been in good health. The pain was over the base of the left big toe. During the year the pain ranged from slight to severe, but during the week previous to attendance it had been extremely severe. She said that the pain became worse on prolonged standing or when playing games, and it was relieved by rest. As a rule the symptoms were absent in the morning. On examination there was swelling and localized tenderness over the plantar surface of the head of the left first metatarsal. There was no sign of infection of the skin. The arches of the feet were normal. There was no hallux rigidus or hallux valgus.

Radiographs showed that the left lateral sesamoid bone was increased in size, deformed, and grossly fragmented

(*Fig.* 761). The bone appeared to be lobulated. The islets of bone were not clearly defined and varied in size. They occupied an extensive area of about 5 times the size of the opposite normal lateral sesamoid. The interspaces between the islets were narrow. Another

FIG. 761.—Radiograph of sesamoid bones of the first metatarsal of both feet showing the gross fragmentation of the left lateral sesamoid bone.

FIG. 763.—Radiograph of the same case taken 10 weeks later showing diminished relative density of the islets.

radiograph taken 4 weeks later showed that the largest islet, which lay between the necks of the first and the second metatarsals, was fragmented and that the small islets were coalesced (*Fig.* 762). A third radiograph taken about 10 weeks after the first attendance showed the relative density of the fragments to be diminished and that the shadow of the islets had begun to become uniform (*Fig.* 763).

Sedimentation rate was 4 mm. in one hour. White blood-cell count and differential count were normal. The temperature was consistently normal.

FIG. 762.—Radiograph of the same bones taken 4 weeks later showing further fragmentation.

The symptoms responded to an insole supporting the arch of the affected foot. In 4 weeks the patient was completely free from symptoms.

After 6 months there was a barely palpable swelling with no tenderness between the heads of the first and the second metatarsals. The patient continued to be free from any symptom.

DISCUSSION

Multiple sesamoid bones may occur normally. If this happens it is the medial sesamoid which is most commonly involved. Fractures of the sesamoid bones are rare. However, the radiographic findings of the apparent density of the bone followed by the porosis that accompanies revascularization, indicates that the primary cause of the case reported here must be avascular necrosis of the bone. The fact that this condition took place at the age of 13, that is at the time of ossification of sesamoid bones, is in favour of osteochondritis. Only one of the other 3 reported cases was an adolescent aged 13 years, the remaining 2 being adults. The fact that in the case described here the fragmentation was greater than in the cases reported by other authors, and that the islets were considerably spread out, may possibly be explained by the fact that the symptoms were present for a year.

I should like to thank Mr. S. Pappworth for permission to report this case.

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CERVICAL TERATOMA

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In 1950 Bale published a comprehensive review of 56 cases of teratoma of the neck, to which he added 4 of his own. In 1952 White and Gosselin reported 1 case and McGoon 4. Two more cases followed their reports, one by Perkins, Pautler, and Winston-Salem (1953), and the other by Hinds, Seybold, and Walker (1954). The first account of a proven case is attributed to Hess (1854). This case was restudied microscopically and confirmed by Wetzel (1895). It is true that two centuries before Hess's report appeared a number of similar tumours had been described, but because no microscopical examination was possible, such reports cannot be accepted as confirmed. Omission of these unconfirmed cases in no way alters the fact that the tumour is rare, for in the hundred years that have passed since Hess's account there have been altogether only 67 cases reported. Two cases of cervical teratoma have been seen at Adeoyo Hospital, Ibadan, in the last 7 years. The first was in a 3-month-old male infant who was under the care of Dr. A. B. Tompkins and Professor Beatrice M. Joly in April, 1953. Histological confirmation of the nature of the tumour was given by the late Dr. W. D. Silvera. The child died a few hours after enucleation of the tumour. The second is the case reported below. Extensive search of the literature has revealed no report of teratoma of the neck in a West African infant; I feel that this, and the fact that the tumour is rare, justify a report of this case.

FIG. 764.—Photograph showing extent of tumour.

FIG. 765.—Appearance 12 days after operation.

CASE REPORT

Foloru A., a Yoruba female infant of 4 months, was admitted to Adeoyo Hospital, Ibadan, on Dec. 26, 1954. Her parents complained of a large swelling on the right side of the child's neck, which they said had been present since birth. The pregnancy was full-term, and birth was normal.

ON EXAMINATION.—A well-nourished child, with a massive swelling which completely occupied the right side of the neck and extended as far as the mastoid process posteriorly and above, producing a forward displacement of the lobe of the ear (*Fig.* 764). The swelling was lobulated, and was partly cystic and partly solid. The skin was freely movable over it, and anteriorly although the swelling was mobile over the deeper structures, posteriorly it seemed attached to the ramus of the mandible. The child had no difficulty in swallowing, and in spite of a slight stridor there was no respiratory distress. No other congenital anomalies were found. Radiographs showed some calcification within the swelling. The tumour was thought to arise from the parotid gland, on account of its apparent attachment to the ramus of the mandible and forward displacement of the lobe of the ear. The alternative diagnosis was a teratoma of the neck.

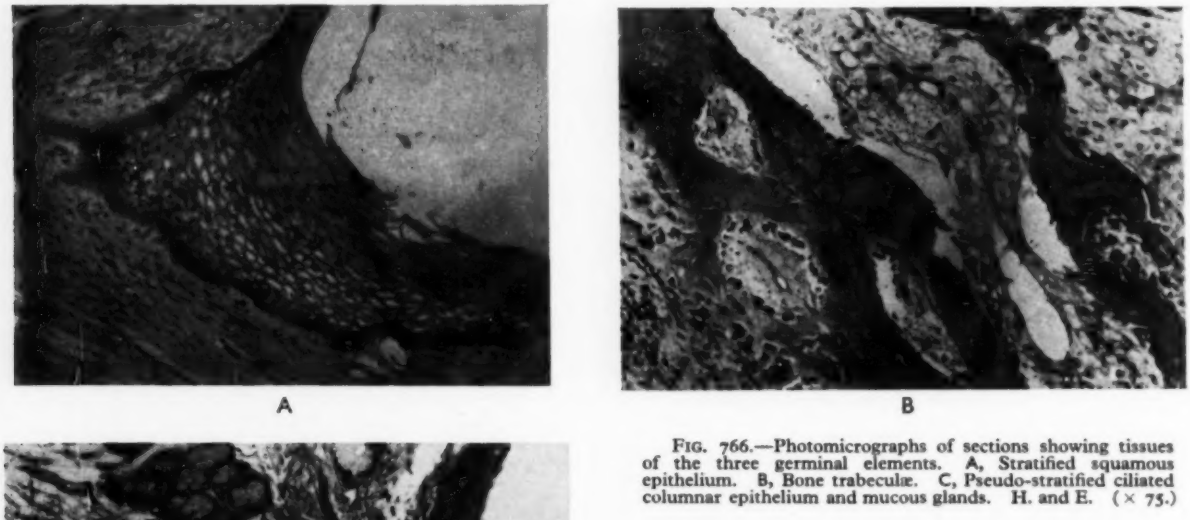
AT OPERATION.—Under general anæsthesia (nitrous oxide with ether) enucleation of the tumour was done on Dec. 29, 1954. This presented no difficulty as it was completely encapsulated. There was no attachment to the parotid or thyroid gland, nor was it supplied with blood by either thyroid artery. The wound was closed over a corrugated rubber drain which was removed in 48 hours. The child made a good recovery and was discharged on Jan. 8, 1955. (*Fig.* 765.) The enucleated specimen was lobulated and roughly spherical, with a diameter of approximately 2·5 in. (6·35 cm.). It possessed a definite capsule, and was loculated on section. Some portions were solid and others cystic. A whitish appearance suggested that the blood-supply was poor.

HISTOLOGICAL REPORT (Professor B. G. T. Elmes).—Nature of specimen: Congenital neck tumour. This is a teratoma. The sections show a variety of tissue arranged in a disorderly manner. All three embryonic layers are represented, but there is a preponderance of

respiratory epithelium, skin, central nervous system, bone, and cartilage. There is no histological evidence of malignancy in any of the components. (*Fig.* 766.)

DISCUSSION

In the pathological classification of teratoma of the neck there seems to be considerable difficulty in determining whether the tumour originates from the thyroid gland or not. Bale (1950) suggested the adoption of three classifications : (1) true teratomas of the thyroid gland, i.e., those tumours which derived their blood-supply from one or other or both thyroid arteries ; (2) teratomas of the neck in the region of the thyroid gland ; and (3) teratomas of the neck probably in the region of the thyroid gland. Classifications (2) and (3) cannot be accepted seriously as pathological distinctions as they do no more than indicate the anatomical position of the tumours. Teratomas classified under (2) had replaced all or part of the thyroid gland, and it was therefore assumed that they grew from embryonic thyroid tissue or from immature tissue in close association with it. Such an assumption is unacceptable until further evidence is obtained to support it. Complete or partial absence of the thyroid gland may be explained equally well by aplasia or hypoplasia and atrophy due to pressure by a large tumour. Similarly, the presence of adult thyroid tissue in a section of the tumour cannot be accepted as irrefutable evidence of thyroid origin, since such tissue may arise from the entodermal component of a teratoma. In the case reported by Perkins, Pautler, and Winston-Salem (1953) additional evidence of thyroid origin of the tumour is claimed because of its attachment to the hyoid bone by tissue resembling the thyroglossal stalk. Histological proof of this claim is not stated. Until a more satisfactory means is found of identifying cervical teratomas originating from the thyroid gland, we must accept Bale's criterion that a common blood-supply to tumour and gland indicates the origin of the former from the latter. If so, then only 6 of the 69 cases so far reported are derived from the thyroid gland.



A B C

FIG. 766.—Photomicrographs of sections showing tissues of the three germinal elements. A, Stratified squamous epithelium. B, Bone trabeculæ. C, Pseudo-stratified ciliated columnar epithelium and mucous glands. H. and E. (× 75.)

A parotid tumour was considered as a possible diagnosis in the case reported here because so-called ' mixed tumours ' of the parotid gland do occur in infancy and early childhood. A tumour related to the angle of the mandible may even be present from birth ; an example was a case of congenital lipoma reported by Willis (1948). Two other differential diagnoses mentioned by authors have been cystic hygroma and congenital goitre. Of the cases reported, 36 were operated on and 31 apparently cured. Of the 5 deaths, 1 occurred in a 41-year-old woman, one month after operation, as a result of metastases. Since this cannot be regarded as death due to operation, we are left with 4 post-operative deaths, which gives an operative mortality of 11 per cent. This good result, considering the age of the patient and the risk of sudden asphyxia if operation is delayed, makes surgical removal of the tumour the treatment of choice. The complete encapsulation seen in most cases facilitates the operation and contributes greatly to its low mortality rate.

SUMMARY

The rarity of cervical teratoma is indicated by the fact that so far only 67 authenticated cases have been reported. The number is brought up to 69 by 2 cases seen in Nigeria, one of which is fully reported. The difficulty of finding a satisfactory classification and of proving the origin of the tumour from the thyroid gland is discussed. Surgical excision is recommended as offering a safe and effective treatment.

Acknowledgements.—I wish to thank Dr. A. B. Tompkins for referring the case to me, and him and Professor Beatrice M. Joly for allowing me to mention their case.

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INNOMINATE ARTERIOVENOUS FISTULA

REPORT OF A CASE

BY J. R. BELCHER, LONDON

ARTERIOVENOUS fistulæ involving the innominate vessels are rare. Hitherto 4 cases have been recorded, and a fifth is presented in this paper.

CASE REPORT

C. W., a man aged 50, was shot at close quarters on Jan. 6, 1956. He received 6 wounds, all of which healed rapidly. One ·22 bullet entered his chest just below the left clavicle and came to rest in the right axilla.

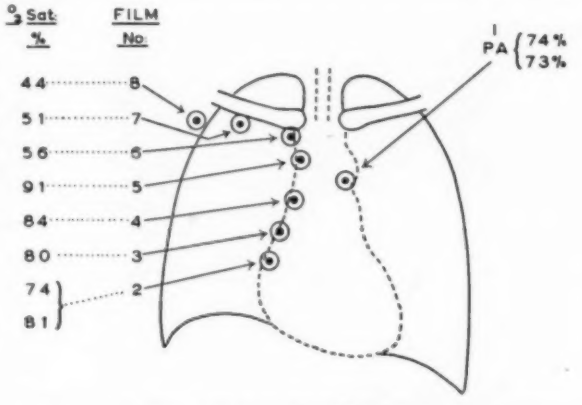

FIG. 767.—A diagram to show the variation in levels of oxygen saturation on cardiac catheterization.

Immediately after this the patient had a small hæmoptysis. Ten days later a diagnosis of an arteriovenous fistula involving his great vessels was made.

He was admitted to the Middlesex Hospital under the care of Mr. R. S. Handley on Feb. 1. His wounds were all well healed and a radiograph revealed a bullet in his right axilla. There was a loud 'machinery' murmur behind the manubrium sterni; this was louder on the right side. His blood-pressure in the right arm was 100/45 and in the left 120/55. His jugulovenous pressure was raised, and his cardiothoracic ratio had increased from 49 per cent to 52 per cent in one month.

Cardiac catheterization was carried out by Dr. Somerville on Feb. 21, and the presence of a fistula was confirmed (*Fig.* 767). From this it was considered that the communication was probably between the right innominate vein and the innominate artery, but from the direction of the wound it was thought more likely that the left innominate vein was involved.

AT OPERATION (Feb. 29, 1956).—A curved incision was made over the front of the sternum, the bone was split from the manubrium to the level of the fourth interspace, and it was divided transversely at this point; good exposure was obtained when the two halves were spread apart. The thymus was removed and a thrill could then be felt over the site of the fistula. The communication was between the anterior surface of the innominate artery and the posterior wall of the left

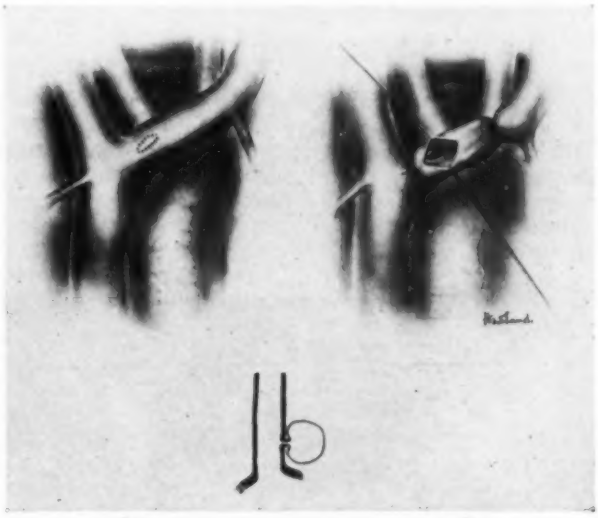

FIG. 768.—A, A drawing to illustrate the site of the fistula between the left innominate vein and the innominate artery. B, To illustrate the appearance of the fistula when the vein had been ligatured and opened.

innominate vein. The orifice, which could be palpated through the vein, was a transverse slit about 1 cm. long. There was a little scar tissue surrounding the fistula, but there was no false aneurysm (*Fig.* 768 A).

The vein was cleaned, and after the superior vena cava had been identified, clamps were applied to it on each side of the fistula. Scarring made taping of the innominate artery appear hazardous, and in any case it was not intended to occlude the vessel even temporarily, unless it proved absolutely essential.

It had been hoped that it would be possible to repair the fistula through the lumen of the vein, but as soon as the clamps were applied the vein became tense and although the fistula felt small it was apparent that there was a large amount of blood passing through it. Three mattress sutures were placed as near as possible to the fistula under the vein; the thrill was thereby almost

abolished. The vein was then ligatured on either side of the fistula in the hope that the risk of recanalization would thus be lessened, and it was opened over the site of the leak. (*Fig.* 768 B.) There was still a considerable flow of blood through the slit, but this could be controlled with a finger. The orifice could be seen clearly and it was sewn with interrupted No. 2/0 Mersilene. All leakage then stopped and the vein was closed over the fistula. The wound was closed with drainage.

There was some œdema of the left hand in the early period after the operation, but this subsided rapidly.

There is now no murmur audible over the site of the fistula, but the left external jugular vein remains prominent. The patient is symptom-free.

DISCUSSION

The treatment of the first 3 recorded cases of innominate arteriovenous fistulæ, those of Lexer (1934), Elkin (1945), and Gordon-Taylor (1950), involved ligature of the artery.

The case recently published by Franklin (1955) was treated in the ideal manner, in that the fistulous track was divided and the two major vessels were separately sutured, but the innominate artery was occluded temporarily during the operation. All the patients concerned were relatively young, the oldest being 37, but as the subject described in this paper was 50 it was thought that partial interruption of the cerebral circulation, even for a short time, should be avoided if possible.

The previous 4 cases were operated upon at periods varying from 4 to 14 months after the injury. In this case it was thought that more urgent treatment was required, and exploration was undertaken 6 weeks after the shooting. From the descriptions of the other operations, surgery may have been rendered easier by this earlier intervention.

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SURGICAL CORRECTION OF THE DEFORMED ARTHRITIC HAND

REPORT OF A CASE

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THE deformities of the rheumatoid arthritic hand are well known. The disability that these people suffer can be appreciated when one observes them trying to cope with the everyday usages to which they put their hands. Despite this it is surprising that so little has been reported in the way of improving the function in the hands of these people; judging from the published cases this is the impression gained. I therefore consider that the following case is worth reporting.

CASE REPORT

The patient, a man aged 48, was referred to me by his doctor, as he was anxious that something should be done to improve the function of his right hand. He gave the history of an attack of rheumatoid arthritis 10 years previously and this left him with a deformity confined to the right hand (*Fig.* 769). There was the typical ulnar deviation of the fingers at the metacarpophalangeal joints. Flexion deformity was severe at these joints, increasing in degree from the forefinger through to the little finger, and extension was severely restricted

FIG. 769.—Photographs of the hand showing the maximum amount of extension possible at the metacarpophalangeal joints. These photographs were taken in the early post-operative period following the first operation, and hence show the early increase in extension permissible at the metacarpophalangeal joint of the little finger.

due to dislocation forwards of the proximal phalanges on the metacarpal heads (*Fig.* 770). The function of the hand was confined virtually to the thumb and forefinger.

It was decided to improve the function of the little finger first.

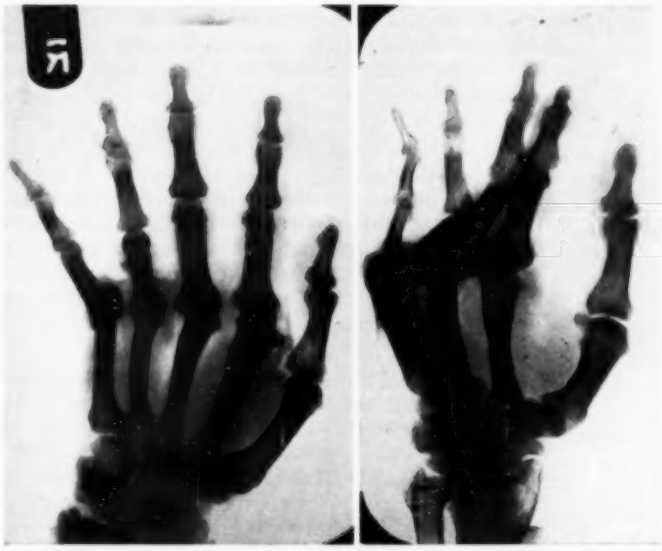

FIG. 770.—Radiograph showing the anterior dislocation of the proximal phalanges on the metacarpal heads.

FIG. 771.—Photograph showing the position of grasp that was finally obtained.

FIG. 772.—The final position of full finger extension achieved by operation.

On April 6, 1955, a longitudinal incision was made over the dorsimedial aspect of the fifth metacarpal. The extensor tendon was found to have slipped over the ulnar side of the shaft and neck of this bone. The head and distal half of the metacarpal were carefully excised, thus permitting full passive extension of the finger. The wound was closed and the digit was splinted in a corrected position. Splintage was maintained for 3 weeks and active movements were allowed. When seen on May 18, active extension of the finger was so good that it was decided to proceed on similar lines with the other deformed fingers. This was done on May 25. The laxity in the tendon of the ring finger was taken up by dividing the extensor tendon, overlapping the ends, and suturing with wire. The middle finger extensor tendon was left alone and the index finger tendon was plicated and sutured with wire.

The same post-operative régime was pursued, and on Aug. 3 physiotherapy was discontinued. *Figs.* 771 and 772 show the full range of movements that were now obtainable. The grip in the hand steadily improved. The patient could now do things that he had not been able to do for years, such as grasping a tumbler, washing his face with the flat of his hand, and easing himself up from a sitting position with the palm of his hand. More recently he has been employed climbing 60-foot ladders in the holds of ships.

FIG. 773.—Radiograph of the right hand to show the resections of the metacarpals.

Fig. 773 shows the radiograph following the resections.

DISCUSSION

Kestler (1946) performed excision of the head and adjacent portion of the metacarpal shaft for the painful arthritic hand, when the pain was confined to the metacarpophalangeal joint, and whether the deformity was present or not. He stressed the undesirability of the interposition of soft tissue and pointed out that it was not necessary to await a normal E.S.R. before operating. In 1950 he reported a further procedure, which amounted to a cartilaginous cup arthroplasty, retaining the cartilaginous cup of the metacarpal following excision of the rest of the head and neck of the bone. This procedure was considered to give more stability to the metacarpophalangeal joints and was thought preferable in the younger age-groups where the cartilage was intact. Steindler (1951) maintained that of all the arthritic deformities those of the fingers had received least attention. He stated that the metacarpophalangeal joints were suitable for arthroplasty and advised excision of the metacarpal heads where there was "irresistible extension contracture of these joints", and particularly where there was dorsal dislocation of the proximal phalanges. Mercer (1954) mentions capsulectomy and metacarpal osteotomy in cases of resistant flexion deformities at the metacarpophalangeal joints. I believe that several centres have tried replacing the metacarpal head by an acrylic prosthesis. No reports on the effectiveness of this procedure seem to be available. On theoretical grounds one would condemn the operation, for with capsular and other soft-tissue contractures being present, a shortening of the metacarpal is the desired aim.

It has been left to Bunnell (1955) to reveal what can be achieved by surgery in the rheumatic hand. Where the dislocation of the metacarpophalangeal joints can be reduced passively he advocates shortening of the metacarpal at the base. Where the dislocation cannot be passively reduced, arthroplasty by excising the metacarpal head is performed. Surgery is only performed on "burnt out cases".

In the case reported here there was marked dislocation at the metacarpophalangeal joints and it was felt that resection of more than the neck of the metacarpal was necessary, depending on the severity of the displacement. The amount of laxity in the extensor tendons following bony resection presented a problem, and in two of the fingers a shortening procedure was carried out. From the final result it would appear that extension was best in just those fingers in which the tendons had not been interfered with. No attempt was made to correct the ulnar deviation apart from post-operative splinting.

If one may be permitted to draw any conclusion from a single case I would say that where there is marked displacement of the proximal phalanx on the metacarpal head a resection of at least the distal third of the metacarpal shaft is required. It would appear to be unnecessary to shorten the extensor tendons. The muscles themselves seem to make the necessary adjustments.

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SHORT NOTES OF RARE OR OBSCURE CASES

GAS GANGRENE SEPTICÆMIA DUE TO CARCINOMA OF THE CÆCUM AND MUSCULAR TRAUMA

By J. C. VALENTINE

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Gas gangrene may follow perforation of the large bowel or appendix, but in such cases the localized nature of the condition is usually evident. Thus in Gordon's (1936) case there was gas gangrene of the right thigh due to an appendix abscess, while in the case described by Wyman (1949) the left thigh was involved from a carcinoma of the descending colon just below the anterior superior iliac spine. The metastatic production of gas gangrene at a distance from such a source must be very rare. Kimball and Rawson (1952) have described 2 cases. In their first case the left arm was involved from a carcinoma of the cæcum, and in their second case the left thigh. No portal of entry was found in this instance. Death was extremely rapid, occurring within twenty-four hours of onset in each case. A somewhat similar case occurred recently at the Bedford General Hospital.

CASE REPORT

A man of 70 had a swelling of the right leg lasting three weeks in 1947. Two years later he had a febrile illness lasting three days, followed by swelling of the left leg below the knee. Diagnosis of femoral thrombosis was made and treated with an elastic bandage. He was next seen in February, 1956, when he was admitted with a history of pain in the abdomen for two days, with vomiting. The pain was of a colicky type, the attacks lasting for about five minutes. On admission, temperature was 99° F., pulse 80/min. There was tenderness of the right iliac fossa where there was a palpable mass. He was thought to have an appendix abscess which was treated conservatively, and he was discharged after nine days to be seen later, with a view to interval appendicectomy. When seen one month later, there was still a small palpable mass, and he was asked to report again in one month. Two weeks later he was brought to the Casualty Department following a fall at home at

6.45 a.m. On the previous day he had complained of pain in the left side of the abdomen with diarrhœa. He had vomited once. After the fall he had complained of pain in the left hip and iliac fossa, and was mentally confused. On admission at midday there was pain over the left hip and right ribs, but no evidence of bony injury. Temperature was 100° F., pulse 76. There was guarding of the right side of the abdomen and a doubtful mass in the right iliac fossa. By 4 p.m. on the same day, there was marked bruising of the left hip with crepitus. An hour later there was considerable extension of the bruising and large blisters filled with blood-stained fluid had appeared on the thigh. Fluid from one of the blisters showed numerous Gram-positive bacilli. At 6 p.m. he died quite suddenly. By this time the left thigh was approximately twice the size of the right.

POST-MORTEM EXAMINATION.—This was started at 11 a.m. the next day, the body having been refrigerated since death. In spite of this, the tissues were bloated due to gas, the body resembling that of a corpse unrefrigerated for several days. There was dusky discoloration of the skin of the left thigh and a smaller area in the right groin, with blisters over the thigh and also on the right side of the chest. There was evidence of recent hæmorrhage into the muscles of the left thigh in the vicinity of the greater trochanter of the femur. The muscle here was grossly necrotic. No fractures were found in the femur or the pelvis. In the cæcum was found a shallow ulcerated area with raised edges—clearly a carcinoma. It was adherent to the posterior abdominal wall, but there was no obviously local infection of the surrounding tissues or direct continuity with the lesions of the left thigh. The appendix was normal. There were gas bubbles in most of the organs, though the brain appeared to be unaffected. The most striking appearance was in the liver, where the right lobe showed numerous gas cysts, the appearances closely resembling those of a rubber sponge. The left lobe, on the other hand, showed no such cysts. Paget's disease of the skull was an incidental finding.

MICROSCOPIC EXAMINATION.—

Cæcum: Well differentiated adenocarcinoma. A few gas cysts in the wall.

Liver: Numerous large gas cysts in the right lobe, a few small gas cysts in the left lobe. Numerous Gram-positive bacilli in both lobes. Cellular detail poorly preserved.

Cardiac muscle: Muscle tissue split up and distorted by numerous gas bubbles.

Bacteriology.—Films were prepared from the heart blood and stained by Gram's method, and similar films were made from a portion of the muscle of the left thigh after grinding it to an emulsion with a little saline in a Griffiths tube. These showed large numbers of Gram-positive spore-bearing organisms.

On culture aerobic blood-plates, both from heart blood and muscle, grew a few colonies of *Bact. coli*, *Strep. fæcalis*, and coagulase-negative micrococci. Anaerobically both produced a culture of Clostridia-like colonies of a uniform morphology. A number of colonies were picked from the culture of each specimen and proved to be identical on further examination. The morphology of the organism and the colonial appearances were consistent with those of *Cl. septicum*. After two days' incubation, colonies on blood-agar were slightly raised, greyish, translucent, with very irregular margins, and surrounded by a zone of hæmolysis. The biochemical behaviour was as follows: cooked-meat medium, gas with slight reddening of the meat; coagulated serum, not digested; gelatin, liquefied; indole-negative; acid and gas produced from glucose, maltose, lactose, and salicin; no change in mannite, sucrose, and glycerol.

On the above characteristics the organism was identified as *Cl. septicum*.

Pathogenicity tests or serum-neutralization tests were not carried out.

DISCUSSION

It is generally believed that the blood in the portal vein exists as two separate streams with little intermingling (Himsworth, 1950). The blood from the superior mesenteric vein is thought to be distributed to the right lobe, while that of the splenic vein goes to the left lobe. If this is so, then any organisms entering the blood-stream in the region of the cæcum would lodge in the right lobe of the liver. The right lobe would then bear the brunt of such an infection. A generalized bacteræmia might subsequently result by escape of organisms into the systemic circulation from the right lobe of the liver.

Something of this sort appears to have happened in the present case. It is almost certain that the bacteria gained entrance via the carcinoma of the cæcum. It may well be that clostridial bacteræmia is not very uncommon, but owing to the anaerobic nature of the organism, no harm results. In fact, Bower, Mengle, and Paxson (1938) showed that a high percentage of persons with infections of the appendix have more than one unit of circulating *Cl. welchii* antitoxin per ml. of blood, while no such antitoxin could be demonstrated in the blood of the controls. In the presence of muscle damage or other cause of lowered oxygen tension, a bacteræmia might result in a localized area of gas gangrene in the damaged tissues. It must remain a matter for speculation as to when the bacteræmia first started in this present case, but it seems likely that the symptoms of the day before the fall may have been a manifestation of it. It may even have made the patient so unwell as to cause him to fall. The muscle damaged in the fall then provided a suitable nidus for the development of gas gangrene and general dissemination of the organisms. Of course, much of the gas found at autopsy was post mortem in origin, but it is difficult to explain the difference between the amounts in the right and left sides of the liver except by postulating some ante-mortem proliferation in this lobe.

That the whole process, from the fall to death, occupied only twelve hours is almost incredible, but the facts do not seem to offer any alternative conclusion. It is a fact, however, that gas gangrene in war injuries may progress with alarming speed and fully developed gas gangrene may be seen only four hours after wounding (Wilson and Miles, 1946; Carling and Ross, 1948). Kimball and Rawson's (1952) first two cases died within twenty-four hours of the onset of the illness.

SUMMARY

A rapidly fatal gas gangrene infection due to *Cl. septicum* is described in a man of 78. The organism gained entrance through a carcinoma of the cæcum, producing a bacteræmia. A fall with injury to muscles of the left hip gave rise to gas gangrene infection of the hip and thigh, followed by general dissemination of the organism.

Acknowledgements.—I am grateful to Mr. R. R. Peatfield and H.M. Coroner for Bedford for

permission to publish this case, and also to Dr. W. Lane, P.H.L.S., for the bacteriological investigations.

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ENTEROGENOUS CYST OF THE STOMACH SIMULATING HYPERTROPHIC PYLORIC STENOSIS

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ENTEROGENOUS cysts are very uncommon and only on rare occasions do they occur in the region of the pylorus. The following case illustrates how a juxtapyloric cyst in an infant may be confused with the tumour of hypertrophic pyloric stenosis.

CASE REPORT

N. C., a baby aged 8 days, was admitted on May 3, 1956, with a history of projectile vomiting after every feed for three days. The child had been born normally at full term and weighed 7½ lb. at birth. She was breast-fed, and for five days her progress had been satisfactory. Meconium had been passed and the bowel actions were normal. She was the third child, the other two, aged 9 and 5 years, being alive and well.

ON EXAMINATION.—A healthy looking infant with a minor degree of dehydration, weight 6 lb. 10 oz. The heart and lungs were normal. Examination of the abdomen revealed visible gastric peristalsis, and on palpation a pyloric tumour could be felt very easily. A diagnosis of hypertrophic pyloric stenosis was made.

AT OPERATION (Mr. P. W. Ingram).—The abdomen was opened under local anæsthesia and a very large pyloric tumour delivered, which was found to be an oval cyst measuring 3 × 2 cm., situated in the wall of the pyloric antrum near the greater curvature.

A general anæsthetic was then administered and the pyloric antrum opened longitudinally. The cyst bulged into the pyloric antrum, narrowing the lumen to a slit. An attempt at enucleation failed because no plane of cleavage could be found between the cyst and the muscle of the stomach wall in which it was embedded. It was therefore aspirated and then removed piecemeal, together with the overlying antral mucosa. The mucosal deficiency thus produced was such that to have sutured it would have caused severe stenosis, and it was therefore ignored and the opening in the stomach sutured transversely, thus ensuring a good lumen, but leaving a diverticulum whose wall consisted of muscular and serosal layers only (*Fig.* 774).

There was some vomiting during the immediate post-operative period, and intravenous Hartmann's solution was administered for two days. Thereafter the infant made an uninterrupted recovery and was discharged on the fourteenth day after operation weighing 6 lb. 12 oz. She continued to thrive, and on Nov. 2, 1956, weighed 21 lb. 8 oz.

PATHOLOGY.—Examination of the specimen removed showed:—

Macroscopical: Three fragments of greyish brown tissue, the largest measuring 2 cm.

Microscopical: This showed fragments of pyloric or antral mucosa and underlying muscle. The latter looked abundant and the individual fibres hypertrophic by adult standards. In the muscularis, just beneath the mucosa, was a cyst lined by connective tissue which was very vascular and showed fairly abundant lymphoid infiltration. The lining membrane was, for the most part, one of modified connective-tissue cells, although there was evidence of flattened epithelium in places. A pale hyaline secretion was present in the lumen.

Contents of cyst: A yellow hazy fluid containing a faint trace of blood, probably due to contamination. Gelatin digestion test for trypsin, negative. Culture, no growth. Protein, 450 mg. per cent. Test for globulin, negative.

DISCUSSION

McNeer and Spitz (1951), who reported a case of a gastric cyst, could find records of only 6 others, whilst Baroody, Innes, and Harrison (1956) gave the total number of duodenal cysts on record as 22.

Enteric cysts of the stomach tend to occur near the pylorus on the greater curvature; they cause obstruction of the narrow lumen, and therefore the majority of cases present in early infancy. The condition is sometimes found in adults, and Peple (1948) describes a pyloric cyst in a man of 69.

Juxtapyloric cysts in infants, whether originating in the duodenum or stomach, produce the clinical picture of hypertrophic pyloric stenosis. One distinguishing feature, however, is the ease with which the tumour can be felt, a fact commented upon by Basman (1938). A cyst presents constant physical characteristics, unlike the transient signs of the tumour of hypertrophic pyloric stenosis.

There appears to be only one previous record of analysis of the contents of a cyst, that of Basman, who found an alkaline fluid containing no amylase, and with a CO_2 combining power of 54·5 vols. per cent.

Morphologically, the main feature of practical importance is the intimate relation of the cyst to its surrounding structures. As Ladd and Gross (1940) point out, it shares a common wall with the intestine and the two cannot easily be dissected apart. This fact has an important bearing on treatment because it makes simple enucleation impossible and accounts for the variety of methods that have been tried. These include: (1) Aspiration; (2) Local excision;

(3) Marsupialization; (4) Excision of the septum between the cyst and the lumen of the bowel; and (5) Gastro-enterostomy.

Basman aspirated a duodenal cyst simulating pyloric stenosis in a 4-weeks-old female child; it

SUMMARY

A case of enterogenous cyst of the stomach simulating hypertrophic pyloric stenosis is described. It was treated by local excision and the infant made a good recovery.

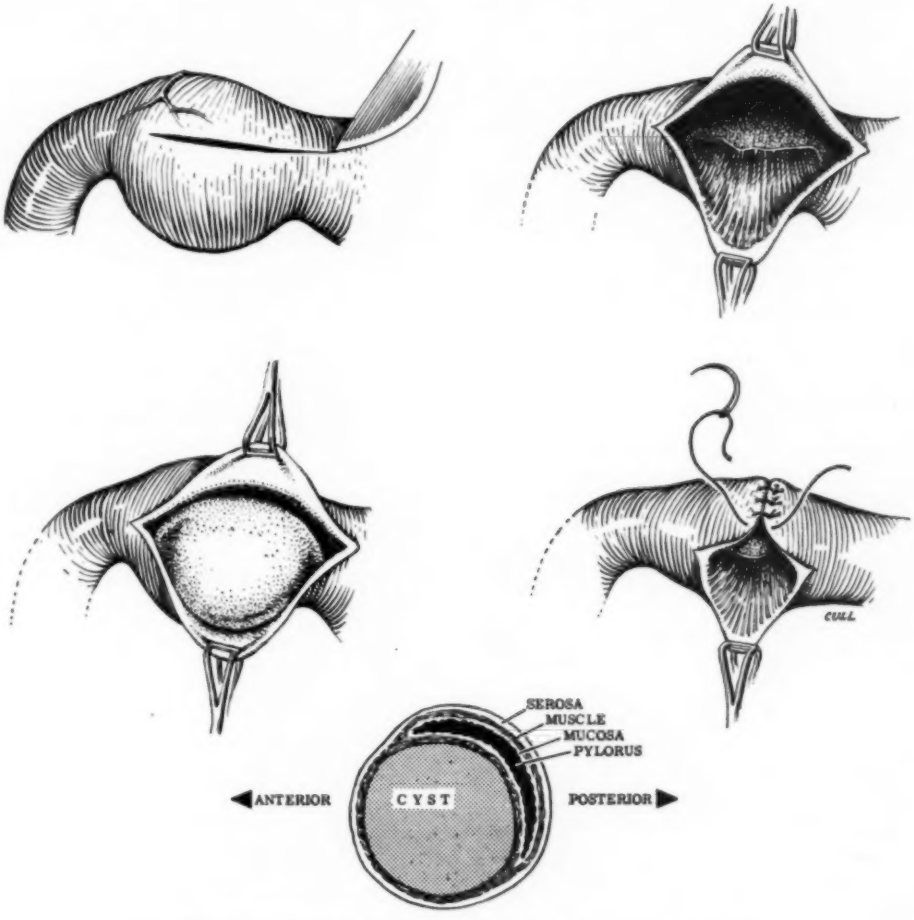

FIG. 774.—Showing the situation of the cyst and the method of excision.

recurred, and at the second operation, a gastro-enterostomy was done, since local excision was found to be impossible.

Smith (1930) reported a similar case in a female child aged 2 weeks; the cyst was drained externally and the child died one week later.

Gordimer and Bluestone (1950) described a cyst in the second part of the duodenum. The patient was a female, aged 30. Part of the septum between the cyst and the lumen of the bowel was removed, thus establishing free drainage.

McNeer and Spitz (1951) advised anterior gastro-enterostomy for a cyst obstructing the pylorus in an infant, with the idea of resecting the lesion later.

Whilst it is obviously impossible to be dogmatic about the treatment of such a rare condition, marsupialization, which according to Gordimer and Bluestone has a mortality of 100 per cent, must be condemned.

I wish to thank Mr. P. W. Ingram and Dr. G. Ormiston for permission to publish this case. My thanks are also due to Dr. H. G. H. Richards for the pathological report and to Mr. Peter Cull for the drawings.

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REVIEWS AND NOTICES OF BOOKS

Training of the Lower Limb Amputee. By DONALD KERR, B.B.A., Director, National Institute for Amputee Rehabilitation, Lodi, New Jersey; and SIGNE BRUNNSTROM, M.A., Consultant in Amputee Training, Institute of Physical Medicine and Rehabilitation, New York University—Bellevue Medical Centre. With an Introduction by T. CAMPBELL THOMPSON, M.D. 9¼ × 6 in. Pp. 272 + xxiv, with 153 illustrations. 1956. Springfield, Ill.: Charles C. Thomas. (Oxford: Blackwell.) 50s.

MR. DONALD KERR is an outstanding exponent of the art of living with an above-knee prosthesis, and Miss Brunnstrom is an international authority on the physical treatment of those who have lost limbs. Their book has been written, *inter alia*, for the amputee himself and is a reflection of the thirst of the American public for detailed information about bodily ailments. Nevertheless, for others to whom it is directed—those concerned with the after-care of amputees—it could well become a standard text-book. Its authority is undoubted. The text is unequivocal yet eminently readable. The illustrations are excellent.

The authors stress the importance of early briefing of the patient and his relatives about the future, and of thorough physical and psychological preparation for limb wearing. The sections on teaching the use of the prosthesis are excellent. Greater stress is placed on pre-walking exercises than is given in this country. Balance and control of movement of the prosthesis are mastered before forward movement is attempted. This must entail rigid self-discipline on the part of the patient. One feels that the stimulus to morale of early ambulation is too useful to waste.

The authors insist that knee-locks and walking-sticks should be unnecessary even for elderly patients. It is doubtful if they have the number of aged patients seen here or indeed whether they would even consider rehabilitation possible in such cases, in which the knee-lock and walking-sticks may be the only means of escape from the wheelchair.

Faulty gait habits and their correction are well described, but the authors in their quest for perfection have not lost sight of the fact that some of us are better endowed by nature than others and that the natural sloucher is unlikely to emulate the natural athlete.

The modifications required for amputations at various levels and for double amputees are painstakingly described. The final chapter is devoted to social and sports activities and, although perhaps coloured by Mr. Kerr's own outstanding achievements in these fields, amputees—particularly the younger ones—will find much to encourage them in these pages.

Achievement Test Cards are shown in an appendix. The index is adequate, but there is no bibliography.

British Surgical Practice: Surgical Progress, 1955. Under the General Editorship of Sir ERNEST ROCK CARLING, LL.D., F.R.C.S., F.R.C.P., F.F.R., Consulting Surgeon, Westminster Hospital; and Sir JAMES PATERSON ROSS, K.C.V.O., M.S., F.R.C.S., F.A.C.S., Surgeon and Director of Surgical Clinical Unit, St. Bartholomew's Hospital; Professor of Surgery, University of London. 9¾ × 6¾ in. Pp. 387 + viii, with 145 illustrations. 1955. London: Butterworth & Co. (Publishers) Ltd. 45s.

THIS 1955 *Progress Volume* is the fifth of an annual series of supplements to the main work, *British Surgical Practice*, in eight volumes, published under the same editors in 1950. " By this means it is hoped to keep the main work up to date in the ever-increasing field of surgical knowledge by original articles, critical surveys, and abstracts."

In addition there is a *Noter-up* section which will guide the reader to all supplementary articles on, or critical surveys of, each subject mentioned in the main work. Every article in the main volumes of the work has a key number. A corresponding key number is on the appropriate page of the ' noter-up ' section so that the reader can study any reference to a subject and information as to the new additional matter which has been incorporated by an article, survey, or abstract since the publication of the main work.

The original articles in this volume cover a wide field, including recent advances in diagnosis such as Abdominal Aortography; Surgical procedures such as Adrenalectomy, and Hindquarter Amputation; and recent developments in Pre- and Post-operative Treatment. These subjects have progressed beyond the experimental stage and are now accepted as standard procedures. The critical surveys lay particular emphasis on the pathology of post-operative repair and convalescence; the assessment of the results in the treatment of carcinoma of the pancreas, the recent advances in pre- and post-operative medication, and early ambulation in the post-operative period. After stimulating the aspiring, and perhaps perspiring young surgeon with such a range of subjects, and considering the proprietary interest which the general public are showing in surgery at the present time, it was very wise of the Editors to include a critical survey of " Developments in the Law in relation to Surgery ". Postgraduate education is absolutely essential for the practice of surgery. This may be very difficult for even the consultant to obtain when working in isolation and therefore Part III, which deals with Abstracts from carefully selected articles which have appeared during the previous year in journals published in English, must be of immense use to a surgeon when considering what is the best procedure he can adopt in a problem case. The reader will be very impressed with the obvious care with which these abstracts have been selected and edited.

The article on Abdominal Aortography by the translumbar route is described and beautifully illustrated with reference mainly to its use in the diagnosis and indication for surgical treatment in renal disease. The method of procedure and complications are given in detail except that of " Dissection of the Aorta " described by Gayles and Laws (*British Medical Journal*, 1956, **2**, 1141). In spite of the possibility of serious complications in a few cases, its value is such that any slight risk should be accepted by the patient; such risk in skilful hands being no more than is inherent in any exploratory operation on a kidney. The descriptions of the technique of Adrenalectomy and the Hindquarter Amputation, each by a universally acknowledged expert, are invaluable for those who are hoping to gain experience. Every surgeon doing abdominal surgery should study the article on Necrotizing Enteritis. This complication is so rarely met with that many surgeons must be quite ignorant of its existence, and yet early diagnosis will enable successful treatment to be instituted. It is generally agreed that the rapidity in the ' turn-over ' of surgical beds is increasing, due to the use of antibiotics, improvements in anæsthesia, the treatment of the shock hæmorrhage syndrome, and other factors. In spite of this, the waiting lists of those requiring operative procedures are increasing, with the tendency to attempt more and more in less and less time. It was written many years ago that " no man by taking thought can add one cubit to his stature "—in other words Nature cannot be hurried. The article on Recuperation after Operation and the critical survey on Repair and Early Ambulation in the Post-operative

Management of General Surgical Patients will well repay careful study and consideration. The Editors and Authors of the various articles, surveys, and abstracts are to be congratulated.

The National Health Service Act (1946) recognized two types of hospitals, the Teaching and Non-teaching. The former was linked with a medical school and placed under a different administration to that of the latter. This distinction has caused an unfortunate misconception, as the Teaching Hospital under the Act refers solely to undergraduate teaching, whereas every hospital with a resident staff is by the terms of the Act an institution where post-graduate teaching should take place and especially so if any of the resident staff are fulfilling the pre-registration year demanded by the Act. It should, therefore, be incumbent upon all Hospital Management Committees to provide the essential facilities for this post-graduate education from the point of view of the Administration; and the Visiting Staff should provide the postgraduate instruction. The Hospital Management Committees could provide library accommodation for the resident staff; it should be comfortable, warm, well illuminated, and quiet. It should contain a few journals and some standard text-books and books of reference. Among the latter such a work as *British Surgical Practice* with its supplementary volumes and the corresponding one on *Medical Practice* would be most useful. To some extent those hospitals which now have difficulties in obtaining resident house officers would find them diminish if attention were given to providing amenities such as a library which could be used by both the resident and visiting staff.

Diseases and Disorders of the Colon. By ANTHONY BASSLER, M.D., F.A.C.P., F.A.C.G., LL.D., Consulting Gastroenterologist, New York Polyclinic Medical School and Hospital; St. Vincent's; Jewish Memorial; Misericordia and Doctors Hospitals, New York City. 9¼ × 6 in. Pp. 217 + xii, with 79 illustrations. 1957. Springfield, Ill.: Charles C. Thomas. (Oxford: Blackwell Scientific Publications.) 51s.

THIS small monograph represents the author's views on colonic diseases and their diagnosis and treatment. Dr. Bassler speaks with authority, for his matured clinical experience of many years has enabled him to write a lucid account of the various conditions.

Although a large section of the book is devoted to parasitic disorders, yet the sections on colonic growths and ulcerative colitis will interest both the surgeon and the physician.

Throughout the book the illustrations are of a high order, especially the radiographs.

A useful little monograph which should prove of value to the student.

A Textbook of Surgical Pathology. By CHARLES F. W. ILLINGWORTH, C.B.E., M.D., Ch.M., F.R.C.S. (Edin.), Regius Professor of Surgery, University of Glasgow; and BRUCE M. DICK, M.B., F.R.C.S. (Edin.), Regional Consultant in Thoracic Surgery, Western Region, Scotland. Seventh Edition. 9½ × 6¼ in. Pp. 730 + viii, with 322 illustrations. 1956. London: J. & A. Churchill Ltd. 63s.

IN 1932, when the first edition of this well-known volume appeared, we predicted that it would take its place as one of the really useful books for the medical student and practitioner. Since that time six editions have appeared and several of them have been reprinted. This is quite enough to convince the critical reviewer of the usefulness of this work. It has been said that surgeons should not teach surgical pathology and that pathologists should undertake this work. We consider that it would be fatal to the teachings of undergraduate student if ever such a step were contemplated. Surgical pathology is the pathology of the living and as such should be undertaken by practical and experienced surgeons. The seventh edition of this work bears out these statements and no better book on surgical pathology has so far been published. The matured clinical background of the authors is manifested in the lucid descriptions in the text.

The illustrations are a great feature of this book; they are universally good and enhance the value of the written word.

Surgeons as a whole will find this book an excellent reference volume.

Operative Surgery. Edited by CHARLES ROB, M.C., M. Chir., F.R.C.S., Professor of Surgery, St. Mary's Hospital, London; and RODNEY SMITH, M.S., F.R.C.S., Surgeon, St. Georges Hospital, London. Volume 3. 11¼ × 8¾ in. Part IV: Rectum and Anus. Pp. 1–96. Part V: Thorax. Pp. 1–215 + xvi, with numerous illustrations. 1957. London: Butterworth & Co. (Publishers) Ltd. £5 10s. per volume.

THE third volume of this work is devoted to the various operative procedures and techniques that are in use in the surgical treatment of disease of the rectum, anus, and thorax.

The outstanding characteristic of the three volumes of this work which have been published so far is the enormous number of excellent drawings which depict the various stages of the various operations, and the vast majority of these illustrations are original.

There are some twenty-four contributors to this volume and they have all acquitted themselves well, and have produced a work of which they all may be proud.

No surgeon at home or abroad can afford to be without this great work on his bookshelf.

Atlas of Tumours of the Nervous System. By H. M. ZIMMERMAN, M.D., Chief, Laboratory Division, Montefiore Hospital, Professor of Pathology, Columbia University, New York City; MARTIN G. NETSKY, M.D., Associate Neuropathologist, Montefiore Hospital, New York City; and LEO M. DAVIDOFF, M.D., Professor and Chairman, Department of Surgery, Albert Einstein College of Medicine, New York City. 10¾ × 7¾ in. Pp. 191, with 277 illustrations, 223 in colour. 1957. London: Henry Kimpton. £9.

THIS is a beautifully illustrated atlas devoted to tumours of the nervous system. The vast majority of the illustrations are in colour and the photomicrographs are superb.

This volume will be of real value not only to neurosurgeons and pathologists, but also to neurologists for the profusion of illustrations makes this a reliable book of reference.

Surgery of the Anus, Anal Canal and Rectum. By E. S. R. HUGHES, M.D. (Melbourne), F.R.C.S. (Eng.), F.R.A.C.S., Surgeon to Out-patients, The Royal Melbourne Hospital, Melbourne, Australia. With a Foreword by C. NAUNTON MORGAN, M.S., F.R.C.S., Surgeon St. Bartholomew's Hospital, London. 10 × 7 in. Pp. 304 + xii, with 276 illustrations. 1957. Edinburgh: E. & S. Livingstone Ltd. 50s.

ALTHOUGH there are many and varied books devoted to the surgery of the rectum there is always room for new ones provided they have something fresh to tell the reader.

Mr. Hughes has presented us with an admirable monograph based mainly on his personal experience.

Having had the advantage of being nurtured at St. Mark's Hospital, he has the right background to make progress in his speciality. We would like to congratulate the author on the excellence and clarity of the illustrations in this book, especially the line drawings, the majority of which are original.

It is refreshing to see a pleasing picture of Dr. Cuthbert Dukes opposite the title page, for his researches into the pathology of rectal growths have been of the greatest use to proctologists and surgeons in general.

We heartily agree with Mr. Naunton Morgan, who writes a foreword to this book: " The book is up to date, concise, full of practical facts, free from verbosity, and garnished by historical illustrations ".

These are the facts.

Injuries of the Hand. By RONALD FURLONG, F.R.C.S., Assistant Orthopædic Surgeon, St. Thomas's Hospital; Orthopædic Surgeon, Rowley Bristow Hospital, Pyrford and Queen Victoria Hospital, East Grinstead. 10 × 6½ in. Pp. 215, with 99 illustrations. 1957. London: J. & A. Churchill Ltd. 36s.

THIS is a useful practical monograph covering the treatment of hand injuries. In this traumatic age hand injuries are on the increase and the results of treatment are much better than they were a decade ago; this is probably due to the introduction of hand clinics in most of our large hospitals, together with the universal use of the antibiotics.

This monograph is written in an easy lucid style, it is well illustrated, and should prove of real value to resident surgeons, registrars, and house officers, while senior medical students will find much of interest within its pages.

The Cytology of Effusions in the Pleural, Pericardial and Peritoneal Cavities. By A. I. SPRIGGS, D.M.(Oxon), M.R.C.P., with coloured drawings by GERETH M. SPRIGGS. 10 × 7¼ in. Pp. 71, with 40 illustrations, several in colour. 1957. London: Heinemann (Medical Books) Ltd. 42s.

CYTOLOGY has come to the fore in recent years, especially in the diagnosis of malignant conditions. This beautifully illustrated monograph gives a comprehensive account of the cells found in serous fluids in human diseases. Full details are given of the various techniques which are required for making satisfactory films.

The coloured plates are excellent and should prove of value to all who are interested in the pathology of the cell.

Although of essential value to the clinical pathologist, yet all workers in the field of cancer will find this book of considerable value.

Outline of Fractures, including Joint Injuries. By JOHN CRAWFORD ADAMS, M.D., F.R.C.S., Consultant Orthopædic Surgeon, St. Mary's Hospital, London, and St. Vincent's Orthopædic Hospital, Pinner. 8¾ × 5½ in. Pp. 248 + viii, with 218 illustrations. 1957. Edinburgh: E. & S. Livingstone Ltd. 27s. 6d.

THIS is an excellent book. It is primarily written for medical students and physiotherapists, but could be usefully employed in the teaching of orthopædic nurses. With this aim in view the author has been purposefully dogmatic and has been able to present a very clear résumé of established fracture treatment. The summaries at the end of the two main sections of the book should prove very useful to the student who is shortly to be faced with the ordeal of a finals *vive*. The historical footnotes on surgeon's names associated with fractures should also be valuable to the examinee. An orthopædic surgeon is bound to differ with the author on certain points but the differences are largely ones of personal stress on a particular form of treatment, and in most instances mention has always been made of other methods. Mr. Adams is to be congratulated on producing this valuable addition to the student's armamentarium.

Orthopædic Nursing. By MARY POWELL, S.N.R., M.C.S.P., Orthopædic Nursing Certificate, Matron, Nuffield Orthopædic Centre, Oxford. With a Foreword by SIR REGINALD WATSON-JONES, B.Sc., M.Ch.Orth., F.R.C.S., F.R.C.S.Edin.(Hon.), F.R.A.C.S. (Hon.), F.A.C.S. (Hon.). Second edition. 8¾ × 5½ in. Pp. 440 + xii, with 271 illustrations. 1956. Edinburgh: E. & S. Livingstone Ltd. 27s. 6d.

THIS book was first published in 1951 and was written " primarily for nurses and physiotherapists working in orthopædic hospitals while training for the Orthopædic Nursing Certificate ". It was dedicated to the " memory of Dame Agnes Hunt and to the student nurses of the hospital which still bears her name ". This was the Shropshire Orthopædic Hospital, Oswestry, but the name was changed by the Board of Management shortly after the death of Sir Robert Jones, on January 18, 1933, to the "Robert Jones and Agnes Hunt Orthopædic Hospital".

In the preface to the first edition the author stated that " it is based on the teachings of our great founder, Dame Agnes Hunt, the pioneer of orthopædic nursing and herself the victim of a crippling disease ".

Agnes Hunt maintained that proper treatment of the orthopædic patient lies not only in the care of the body but in the creation of an atmosphere of hope, of happiness, of security, and of homeliness. This was enhanced when in 1908 The Board of Education arranged that a long stay orthopædic hospital should be combined with a Residential School and specially selected teachers were to be supplied by the local Education Authority. The first to be inaugurated was the Lord Mayor Treloar Cripples Hospital at Alton associated with the name of Sir Henry Gauvain. Similar schools were inaugurated at other long stay hospitals such as the Wingfield Morris hospital in Oxford and the King Edward VII Hospital under the Sheffield Municipal Authority. Provision had already been made for the education of defective and epileptic children by the Elementary Education Act of 1899. In her opinion these considerations were of infinitely more importance than tidy wards and tidy beds. In the preface to this the second edition the author maintains, and few will disagree with her, that rest, both general and local, fresh air, and good food and occupation are of more importance in orthopædic nursing than antibiotics. Who was this Dame Agnes Hunt? She was a member of an old Shropshire family; her brother Rowland was for a time M.P. for the Ludlow division of Shropshire. She was a cripple herself from early years due to osteomyelitis of the femur. She entered the Royal Alexandra Hospital, Rhyl, as a student nurse in 1887 and completed her training at the Salop Infirmary in 1890 and joined the Queen's Jubilee Nurses in 1891. She and her friend, Miss Goodford (Goodie), took over a derelict house in Baschurch near Oswestry and decided to nurse there, as an experiment, convalescent children suffering from tuberculous infection of joints which in those days often needed multiple operative procedures to eradicate the disease or to correct the deformity. At first the children were admitted only from the Salop Infirmary. A Committee was formed and on October 1, 1900, the house was opened as a hospital to receive four little girls and four little boys needing country air and good food. The accommodation in the house being quite unsuitable for nursing cripples, Miss Hunt arranged that some sheds, which were being used as stables, should be taken from her brother's nearby estate, and, with the aid of a local

handyman had them re-erected in her garden as small wards; each had a floor, roof, and three sides, the remaining side being permanently open to the air. The children lived in these makeshift open-air wards night and day all the year round. Thus the first really "open air" hospital came into being in this country. Since then "open air" hospitals have been built in many parts of this country, the Dominions, the United States, and other parts of the world.

It is impossible to think and estimate the work of Agnes Hunt without linking it with that of Sir Robert Jones, to whom from time to time she would take a bevy of her patients to his out-patient clinic at the Royal Southern Hospital in Liverpool. Their first informal meeting has been described by Sir Robert himself: "My first experience of Miss Hunt" he said "occurred in the Out-patient Department at the Royal Southern Hospital when I saw a lady on crutches in charge of six children lying upon a railway barrow. After a conversation with her, not knowing she was by my side, I said to my House Surgeon 'This is a very intelligent woman'. She heard the observation and I shall never forget her ringing laugh and the reply 'you bet!'". The late Mr. McCrae Atkin has also described how, when he was House Surgeon to Sir Robert Jones, Agnes Hunt would arrive at the out-patient department with usually eight crippled children on a home-made handcart, which had been wheeled from the station by an outside porter; this was a cheaper method of transport than that in use to-day in the Welfare State. Mr. McCrae Atkin, recalling his recollection of the Baschurch Hospital in later years said "The impression that remains is that it was impossible to believe that such crude huts and buildings could be a hospital, but the chief memory is of the woman who made it, who proved that it is the work done that counts, and that it *can* be done without waiting for elaborate and expensive equipment". Can it be wondered that the author dedicates her book to Agnes Hunt?

After the first informal meeting, between these two pioneers, the surgeon in 1904 accepted the invitation to attend at the Baschurch Hospital once a month. Agnes Hunt has described a typical day in their work: "He would arrive at 10.30, at eleven o'clock work began and from forty to fifty out- and in-patients were seen. At one o'clock operations started and generally lasted till six o'clock; then a meal and he returned to Liverpool".

Sir Robert Jones in 1925 said: "Over twenty years ago, with the help of Miss Hunt we started the first really open-air hospital in the world where the children lived in the open air by day and night all the year round". "The sheds were cheap and decrepit—there was an air of penury about it all—but it was the birthplace of a great reform which is steadily gaining ground all over the world". What of the book under review? As one would expect, born of such inspiration it contains in orderly sequence, with beautiful illustrations, all that a nurse can adequately absorb as an introduction to orthopædic nursing.

The importance of orthopædic surgery, coupled as it now is with traumatic surgery in large centres, is very obvious. It is the most logical type of surgery, dealing as it does with deformity both congenital and acquired, as the result of injury or disease. It, together with its sister surgical specialty, named plastic surgery, covers the prevention and repair of deformity and the healing and repair of wounds in the broadest sense. The responsibilities of those practising and teaching it are very onerous as it deals primarily with the principles which govern the practice and scientific development of all the surgical specialties. In other words, the practitioners of these branches of surgery are those who should be designated "general surgeons". All who read the book will realize that the orthopædic nurse and the patient form the pivot round which the whole team and work revolves.

The format and decor of the book is such as is now expected from the publishers. It would enhance its value to successive generations of orthopædic nurses if a photograph of Dame Agnes Hunt could be included in subsequent editions, and a short description of what she did. A footnote should also be included stating that the orthopædic nurse who wishes to realize the tradition she inherits should read *This is my Life* by Agnes Hunt, D.B.E., R.R.C., published in 1938 by Messrs. Blackie & Son Ltd., and *The Life of Sir Robert Jones*, by Frederick Watson, his son-in-law, published in 1934 by Messrs. Hodder & Stoughton Ltd.

Les Ictères par Retention. By J. CAROLI, in collaboration with A. CHARBONNIER, J. EPEVE, A. PARAF, and R. RICORDEAU. 9⅞ × 6¾ in. Pp. 482, with 308 illustrations. 1957. Paris: Masson. Paper covered, 6000 Fr., Cloth bound, 6700 Fr.

OBSTRUCTIVE jaundice is a condition in which exact diagnosis before operation is often difficult and speculative. It may even remain in doubt when the abdomen is open, and the surgeon's best course is often by no means clear. The object of Dr. Caroli's book is to show that a precise anatomical and pathological diagnosis is possible. The value and technique of accepted methods of investigation are fully reviewed, but the main purpose of the book, for surgeons, is to demonstrate the method of radio-manometry of the common bile-duct which has been developed by Dr. Caroli and the French school of surgeons. At operation the unopened duct is injected with radio-opaque material and the pressure is increased until the solution just begins to escape from the papilla into the duodenum. Dr. Caroli shows how this information can help in difficult cases. Perhaps the most important application of the method is in the diagnosis of carcinoma of the distal end of the common bile-duct proximal to the ampulla. The book has some beautiful illustrations to show how a diagnosis between carcinoma, calculus, stenosis, or spasm can be made with confidence by means of radio-manometry.

Clinical aspects of obstructive jaundice are discussed with a critical assessment of the value of the different items, in the light of the author's special interest in the condition over twenty years, and more than 500 cases.

The French style is clear and direct and the book is delightfully produced with clear print on good paper and a wealth of pictures, diagrams, and the author's wisdom distilled in his characteristic crisp sentences. Dr. Caroli's book will have an important effect on surgical practice, emphasizing as it does the need to bring radiological methods into the operating theatre. The book should be read by all surgeons who wish to be fully equipped to deal with a case of obstructive jaundice.

Recent Advances in Anæsthesia and Analgesia—Including Oxygen Therapy. By C. LANGTON HEWER, M.B., B.S., M.R.C.P., F.F.A.R.C.S. (Eng.), Anæsthetist in charge of Department, St. Bartholomew's Hospital, London; and J. ALFRED LEE, M.R.C.S., L.R.C.P., F.F.A.R.C.S. (Eng.), Consultant Anæsthetist to the Southend-on-Sea Hospital. Eighth Edition. 8¼ × 5¼ in. Pp. 295 + viii, with 95 illustrations. 1957. London: J. & A. Churchill. 40s.

TWENTY-FIVE years have elapsed since this admirable book was first introduced to the specialty of anæsthesia, and during this time it has acquired a great reputation for its efficiency in describing recent advances in anæsthesia and analgesia. We venture to assert that the new, eighth, edition is the best of the whole series because the book now only contains developments and advances in anæsthesia during the past few years. This has enabled the size of the book to be kept to reasonable proportions.

The original author, Dr. C. Langton Hewer, has now been joined by Dr. J. Alfred Lee, and their joint effort admirably covers the whole range of recent advances. Of the many excellent chapters, we especially recommend post-graduates to read those upon Muscle Relaxants, Induced Hypotension, Anæsthetic Explosions, and Phenothiazine Derivitives.

We must congratulate the publishers upon the excellent arrangement and production of this book. The illustrations are numerous and excellent, and the printing is very clear. We recommend this book to all post-graduate anæsthetists, especially those studying for the higher examinations in anæsthesia. In conclusion, we would also mention that many surgeons may well be interested in the chapter on Oxygen Therapy.

Cleft Lip and Palate. By W. G. HOLDSWORTH, F.R.C.S., Surgeon, Rooksdown House, Basingstoke; Consultant in Plastic Surgery, South-West Metropolitan Hospital Region. With a Foreword by Sir HAROLD GILLIES, C.B.E., F.R.C.S. Second edition. 10 × 7¼ in. Pp. 187 + xii, with 146 illustrations. 1957. London: Heinemann (Medical Books) Ltd. 42s.

IT is just over six years since the first edition of this monograph appeared, and this second edition covers all the advances that have been made in the subject in recent years.

The author is to be congratulated on the inclusion of an interesting chapter on the embryology, development, and growth of the face contributed by Professor Richard Harrison. Another chapter on orthodontic treatment by Mr. M. A. Kettle enhances the usefulness of the book. Many new and original illustrations are included in this new edition.

Principles and Methods of Sterilization. By JOHN J. PERKINS, M.S., Director of Research, American Sterilizer Company, Erie, Pennsylvania. 9¾ × 6¾ in. Pp. 340 + xii, with 174 illustrations. 1956. Springfield, Ill.: Charles C. Thomas. (Oxford: Blackwell Scientific Publications.) 60s.

"FOR it is not necessary, as Roger and Roland have written, as many of their disciples teach, and as all modern surgeons profess, that pus should be generated in wounds. No error can be greater than this, such a practice is indeed to hinder nature, to prolong disease, and to prevent the conglutination and consolidation of the wound."—Henri de Mondeville (1260–1320).

The prevention and treatment of wound sepsis is no new subject. One can only agree with the publishers when they state on the jacket that this book should be of interest to Hospital Administrators, Physicians and Surgeons, Operating Room and Central Service Supervisors, Directors of Nursing, Clinical Instructors, Laboratory Technicians, Pathologists and Bacteriologists, and Maintenance Engineers. It is extraordinary that they do not mention the calling of the Hospital Architects, who as a profession are difficult to convince on some detail in a building concerning which they are certain they know the last word, but which they do not have to test in actual practice. The author uses the word "standardize" on many occasions but can anything in the practice of medicine and surgery be really "standardized" when the main person involved is one, who also is not mentioned, and that is the human patient.

One can also agree with the author when he states that "The mode of life of modern men is profoundly influenced by hygiene and medicine, and the principles resulting from the discoveries of Pasteur". It was the discovery by Pasteur of the fundamental cause of sepsis and the practical application by Lister that established antiseptic surgery. The terms *antisepsis* and *antiseptic* had been in use for a century before Lister to describe the principle of treatment by various substances, amongst which were carbolic acid and iodine. Lister, in his first paper describing the new antiseptic treatment of wounds (*Lancet*, March 16, 1867), when referring to the question whether the atmosphere produced the decomposition of organic substances, stated that "a flood of light had been thrown upon this important subject by the philosophic writings of M. Pasteur" and that the doctrines described in those writings had been the foundation of the net method of treatment. It was in 1890 at the International Medical Congress in Berlin that von Bergmann, Professor of Surgery at the University Clinic, invited Lister to see several cases of amputation of the breast in which the new aseptic *methods and technique* had been employed; "when the dressings were removed pus was revealed in every case". It has been emphasized that "Lister's invention is the greatest that has been made in the domain of surgery". The only criticism or what some have even regarded as a fault of it, is, that by it and the evolution of aseptic techniques many operators have been created who are not surgeons. With the help of antiseptics, aseptic methods, and antibiotics operative procedures can now be undertaken with safety, almost impunity, but it does not follow that there are indications for their performance.

It is with thoughts such as these that this book should be read and studied by all who work in hospitals, and especially so at the present time when new hospitals are being built and old ones modernized. The ideas of the planners and builders should be determined by the slogan which permeates the book—namely, "Efficiency, economy, and safety", but there must be no excuse for attaining the two former at the expense and sacrifice of the latter. The various chapters follow each other in orderly sequence, beginning with a meagre reference to the historical background of the subject, passing thence to a consideration of the principles and the various methods of sterilization which can be used. There are chapters on the preparation, maintenance and sterilization of parenteral solutions, instruments, laboratory appliances, and methods of gaseous sterilization. The most controversial subject is whether a Central Supply Service Department should be instituted in hospitals in preference to the usual departmental methods of sterilization. A Central Service Department is "the service within a hospital which processes, issues, and controls professional supplies and equipment, both sterile and unsterile, to all departments and floors of the hospital for the care and safety of patients, both medical and surgical". When operative procedures were few the surgical nursing staff could fulfil the requirements. The nursing staff still maintain a satisfactory service provided steam for sterilization is available in all theatres, wards, laboratories, operation rooms, etc.

The demand at the present time for sterilized dressings and appliances, etc., is rapidly increasing, and consequently the preparation, laundry, cleaning, maintenance and care of equipment, is occupying more and more of the time of both the trained and student nursing staff, thereby allowing less for the care of the patient. A position will ultimately develop when the supervision of sterilization is everybody's business and no one's responsibility. The advantages and disadvantages of a central supply department are fully discussed by the author from the point of view of efficiency, economy, and safety. He gives details of the number of staff required for the service, which depends on the number of beds, and includes laundry, repair, drum packing, and storage facilities. Plans drawn to scale are given of the amount of space required, the number of technicians required for the maintenance of equipment and cleaning of appliances such as syringes, instrument trays, assembly sets, etc.

It is obvious, after reading the book, that those planning a hospital should give serious thought as to whether

a central service department is the best solution of their particular problems, paying special attention to finance, accommodation for staff, storage space, which should include offices, rooms for repairs and packing, perhaps a small laboratory, and a room for lecture-demonstrations for instructional purposes. The site for such a department is very important and if not included in the original plan may cause much inconvenience if added later. The supervisor must be one who recognizes the responsibility which rests on all who work in such a department. One of the rarest of human qualities is the ability to look at familiar objects and phenomena with all the interest and curiosity of one who is seeing them for the first time. Ritual tends to become routine unless this ability is kept in mind by all who work in such a department. " Perpetual custom " said Cicero " makes the mind callous and people neither admire nor require a reason for those things which they constantly behold."

The book is one which will repay Hospital Builders for close study.

The opinion and co-operation of the Bacteriologist must be obtained in planning each phase of the building.

Lesions of the Cervical Intervertebral Disc. By R. GLEN SPURLING, M.D., Professor of Neurosurgery, University of Louisville School of Medicine, Louisville, Kentucky. 9½ × 6 in. Pp. 134 + xvi, with 41 illustrations. 1956. Springfield, Ill.: Charles C. Thomas. (Oxford: Blackwell.) 36s.

THIS monograph is by a neurosurgeon already well known in this country as a result of an earlier publication devoted to lesions of the lumbar intervertebral disk.

The present work is an expression of his personal experience with 331 patients with cervical disk lesions, of which 197 cases, 61 of them being subjected to operation, are analysed in some detail.

In the approach to the subject emphasis is laid mainly on the problem of the soft nuclear prolapse as will be seen from the operation findings, for of these 61 cases, 31 (64 per cent) had nuclear herniations, and from this point of view it is in contrast with the attitude taken in this country at the present time where interest tends to be centred about spondylosis of the cervical spine, and which is regarded as the commoner condition.

The book is very simply and attractively written with introductory chapters on the anatomy of the cervical spine and spinal cord, and the pathological changes in the intervertebral disks. There is a great deal of factual information on diagnosis, radiology, and technique of operation which will be of great value to the neurosurgeon.

The production is excellent, with a large number of drawings and photographs of good quality and the book should prove itself to be a valuable addition to the extensive literature on cervical intervertebral disk lesions.

The British Encyclopædia of Medical Practice: Medical Progress, 1956. Editor-in-Chief: Sir HENRY COHEN, M.D., D.Sc., LL.D., F.R.C.P., F.F.R. 9½ × 6½ in. Pp. 380 + xiv. 1956. London: Butterworth & Co. (Publishers) Ltd. 37s. 6d.

" SINCE the last *Medical Progress* volume was published we have lost, through death, its editor-in-chief, Lord Horder, who had been responsible also for second edition of *The British Encyclopædia of Medical Practice*." Thus the new editor-in-chief, Lord Cohen of Birkenhead, begins his editorial article. He then proceeds to pay a tribute to the ability and clinical acumen of the late editor, who " was the first physician to bring the fruits of laboratory and instrumental investigation to the bedside ".

" He remained the trusted doctor to royalty and statesmen, butcher, and beggar, and earned their affection and gratitude." Why and how? because " He never forgot that he was called in to treat a sick man and not simply a disease ". He used to recommend to his students that the first text-book of medicine should be Jevons *Primer of Logic* adding " It costs one shilling but is worth untold gold ". The book is, as its title implies, encyclopædic in the number of subjects with which it deals, and like the similar books published by this firm, in the single basic clinical subjects, it consists of three parts, Critical Survey, Drugs, and Abstracts. The subjects chosen for the surveys are to enable the busy practitioner to maintain as far as possible his postgraduate education. This is difficult owing to isolation, lack of leisure, and incentive. It has to be by reading journals, attending postgraduate courses, lectures, medical society meetings, and now with the help of visual aids such as films or even watching television.

The value of a book such as this is to enable the busy practitioner in whichever branch of medicine his vocation may be, to form some idea as to the extent to which the subject under discussion is at present understood.

The book begins with a brief but comprehensive survey of the year's progress in each of Medicine, Surgery, Obstetrics and Gynæcology, by acknowledged experts.

They are written in such an explanatory manner that they give a much better overall picture of the present state of knowledge of any subject than can be gathered from a prolonged study of the various " Year Books " and " Recent Advances " which are now published, and which are really more useful to the expert than one with a general interest.

The other critical surveys included have been selected with editorial perspicacity, and cover such subjects as blood diseases, chemical pathology, geriatrics, medicine and the law, public health, and radio-active isotopes. These subjects interest not only the general practitioners, but owing to the publicity given to them in the daily press the interest of the man in the street is also aroused and determines him to discuss the subject with his doctor. It is impossible to review such a book in detail; suffice it to say that any medical man will not be wasting his time spending some of his rare leisure moments reading it.

Pulmonary Complications of Abdominal Surgery. By A. R. ANSCOMBE, M.S., F.R.C.S., Senior Lecturer in Surgery and Honorary Consultant, St. George's Hospital. Pp. 121 + x. 1957. London: Lloyd-Luke (Medical Books) Ltd. 20s.

THIS monograph is in reality the 1955 Moynihan Prize Essay awarded by the Association of Surgeons, and there can be little doubt that Lord Moynihan himself would have enjoyed reading the contents of this book.

To-day, with antibiotic cover to abdominal operations, there is a great tendency to neglect post-operative treatment and there are far too many post-operative deaths which could have been prevented.

Mr. Anscombe has given us some interesting facts concerning pulmonary complications following abdominal operations, together with some practical investigations on respiratory physiology.

This book should find its way not only into the library of surgeons but also into the pocket of anæsthetists. Senior students will also find much to interest them in this book, for it is full of interesting clinical facts.

BOOK NOTICES

[The Editorial Committee acknowledge with thanks the receipt of the following volumes. A selection will be made from these for review, precedence being given to new books and to those having the greatest interest to our readers.]

Bone Structure and Metabolism. Edited by G. E. W. WOLSTENHOLME, O.B.E., M.A., M.B., B.Ch.; and CECILIA M. O'CONNOR, B.Sc. 8½ × 5¼ in. Pp. 299 + xii, with 121 illustrations. 1956. London: J. & A. Churchill Ltd. 45s.

General Urology. By DONALD R. SMITH, M.D., Clinical Professor of Urology and Chairman of the Department of Urology, University of California School of Medicine, San Francisco; Consulting Urologist San Francisco Hospital; Consulting Surgeon (Urology), Veterans Hospital, San Francisco. 10 × 6¾ in. Pp. 328, fully illustrated. 1957. Los Altos, California: Lange Medical Publications. $4.50.

Chirurgie de Pancréas. By LUCIEN LÉGER, Assistant Professor of the Faculty of Medicine, Paris; Surgeon of the Paris Hospitals; and JACQUES BRÉHAUT, Assistant Professor of the Faculty of Medicine, Algiers. 9¾ × 6½ in. Pp. 504 + iv, with 154 illustrations. 1957. Paris: Masson. Paper covered 4,200 fr. Board covered 4,800 fr.

La Gastrectomie pour Ulcère. By R. de VARNEJOUL, Professor of the Faculty of Medicine, Marseilles; E. HENRY and R. DEVIN, Assistant Professors of the Faculty of Surgery, Marseilles. 9¾ × 6½ in. Pp. 210, with 96 illustrations. 1957. Paris: Masson. Paper covered 2,400 fr., Board covered 3,000 fr.

Halsted of Johns Hopkins. The Man and His Men. By SAMUEL JAMES COWIE, M.D., Professor Emeritus of Laryngology and Otology, The Johns Hopkins University, Baltimore, Maryland, U.S.A. 9¼ × 6 in. Pp. 247 + xii, with 6 illustrations. 1957. Springfield, Ill.: Charles C. Thomas. (Oxford: Blackwell Scientific Publications Ltd.) 37s. 6d.

Suprapubic Closure of Vesicovaginal Fistula. By VINCENT J. O'CONNOR, B.S., M.D., F.A.C.S., Professor and Head of Department of Urology, Northwestern University Medical School, Chicago, Illinois. 8¾ × 5½ in. Pp. 53 + xii, with 14 illustrations. 1957. Springfield, Ill.: Charles C. Thomas. (Oxford: Blackwell Scientific Publications Ltd.) £1.

Textbook of Orthopædic Medicine. Volume I. Diagnosis of Soft Tissue Lesions. By JAMES CYRIAX, M.D. (Cantab.), M.R.C.P. (Lond.), Physician to Department of Physical Medicine, St. Thomas's Hospital, London. 8¾ × 5¾ in. Pp. 711 + xiv, with 131 illustrations. 1957. London: Cassell & Co. Ltd. 45s.

A Manual of Human Anatomy. Volume III: Abdomen and Pelvis. By J. T. AITKEN, M.D., Reader in Anatomy, University College, London; G. CAUSEY, M.B., F.R.C.S., SIR WILLIAM COLLINS, Professor in Anatomy, Royal College of Surgeons, London; J. JOSEPH, M.D., M.R.C.O.G., Reader in Anatomy, Guy's Hospital Medical School, London; and J. Z. YOUNG, M.A., F.R.S., Professor of Anatomy, University College, London. 8½ × 5½ in. Pp. 135 + viii, with 29 illustrations. 1957. Edinburgh: E. & S. Livingstone Ltd. 12s. 6d.

A Manual of Human Anatomy. Volume V: Central Nervous System. By J. T. AITKEN, M.D., Reader in Anatomy, University College, London; D. A. SHOLL, B.Sc., Reader in Anatomy, University College, London; and J. Z. YOUNG, M.A., F.R.S., Professor of Anatomy, University College, London. 8½ × 5½ in. Pp. 140 + viii, with 40 illustrations. 1957. Edinburgh: E. & S. Livingstone Ltd. 12s. 6d.

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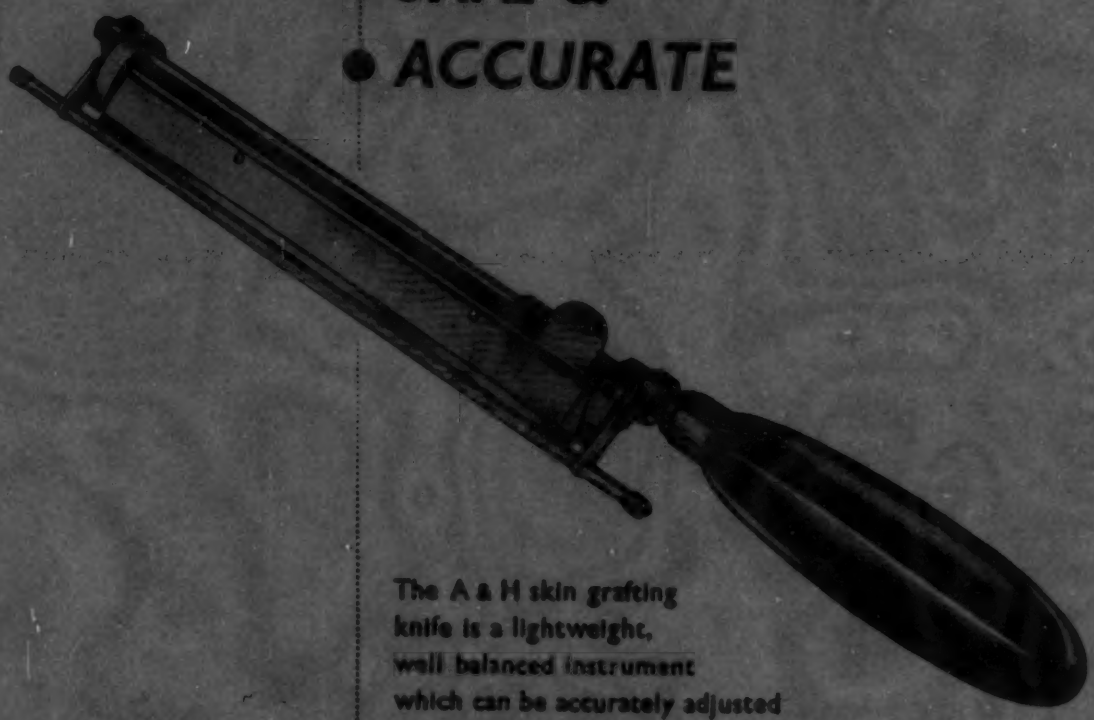

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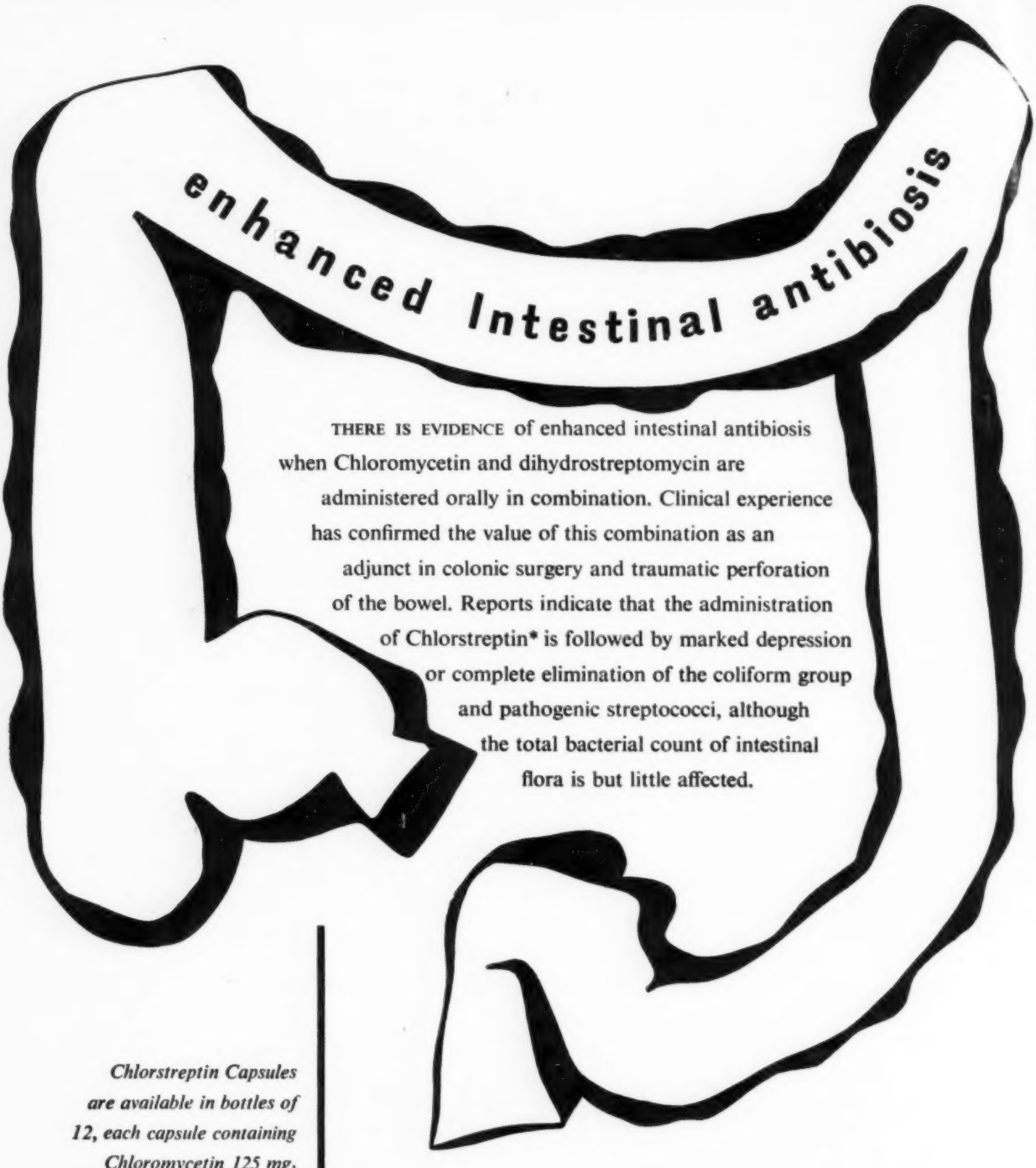

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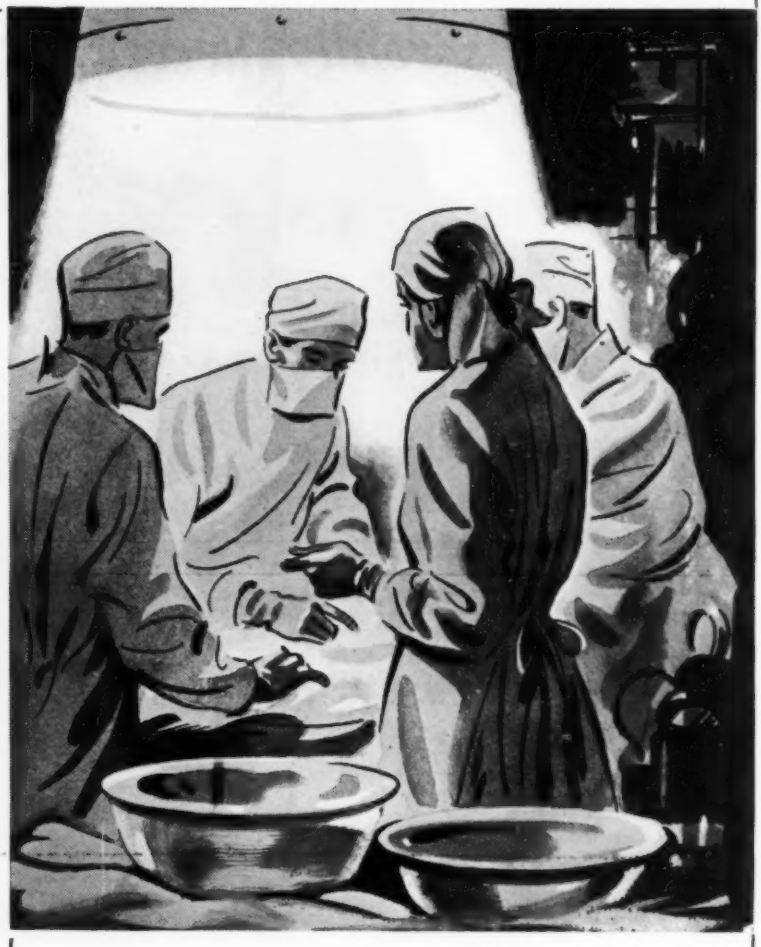

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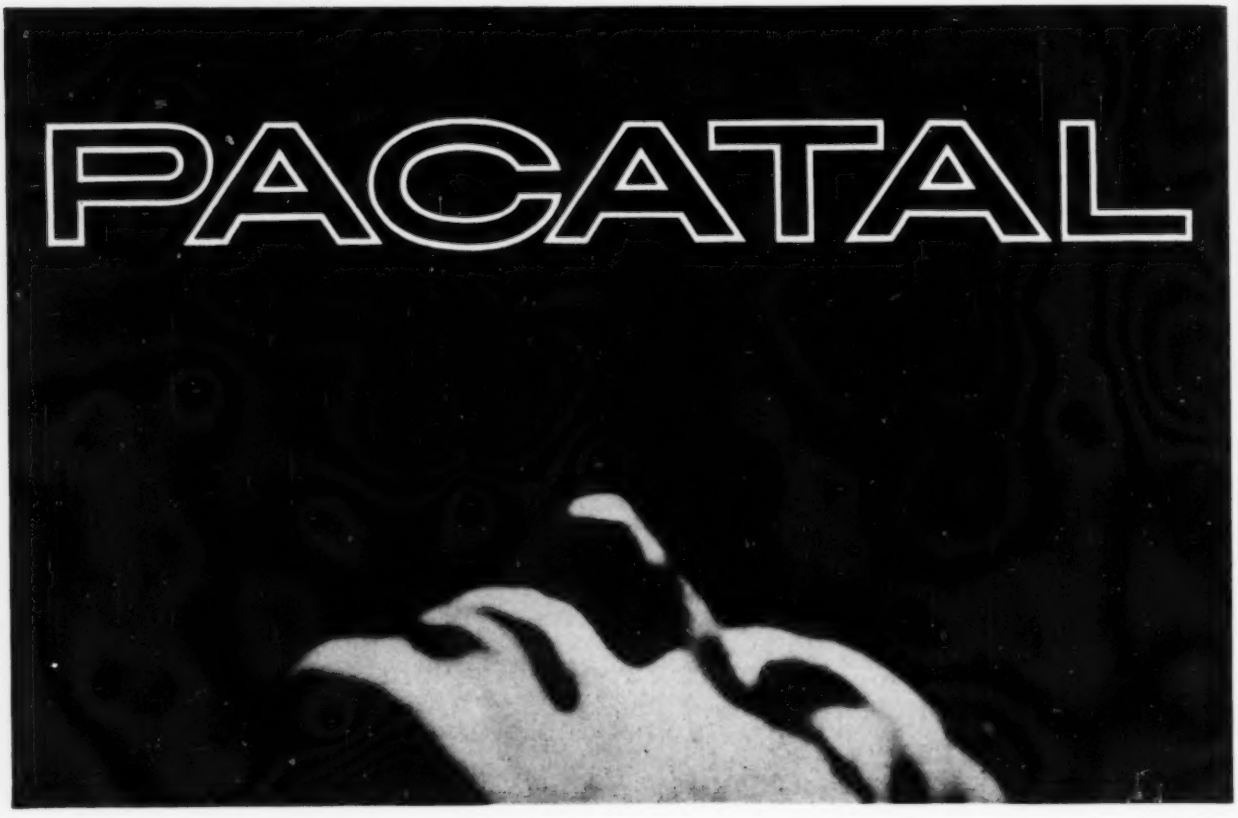

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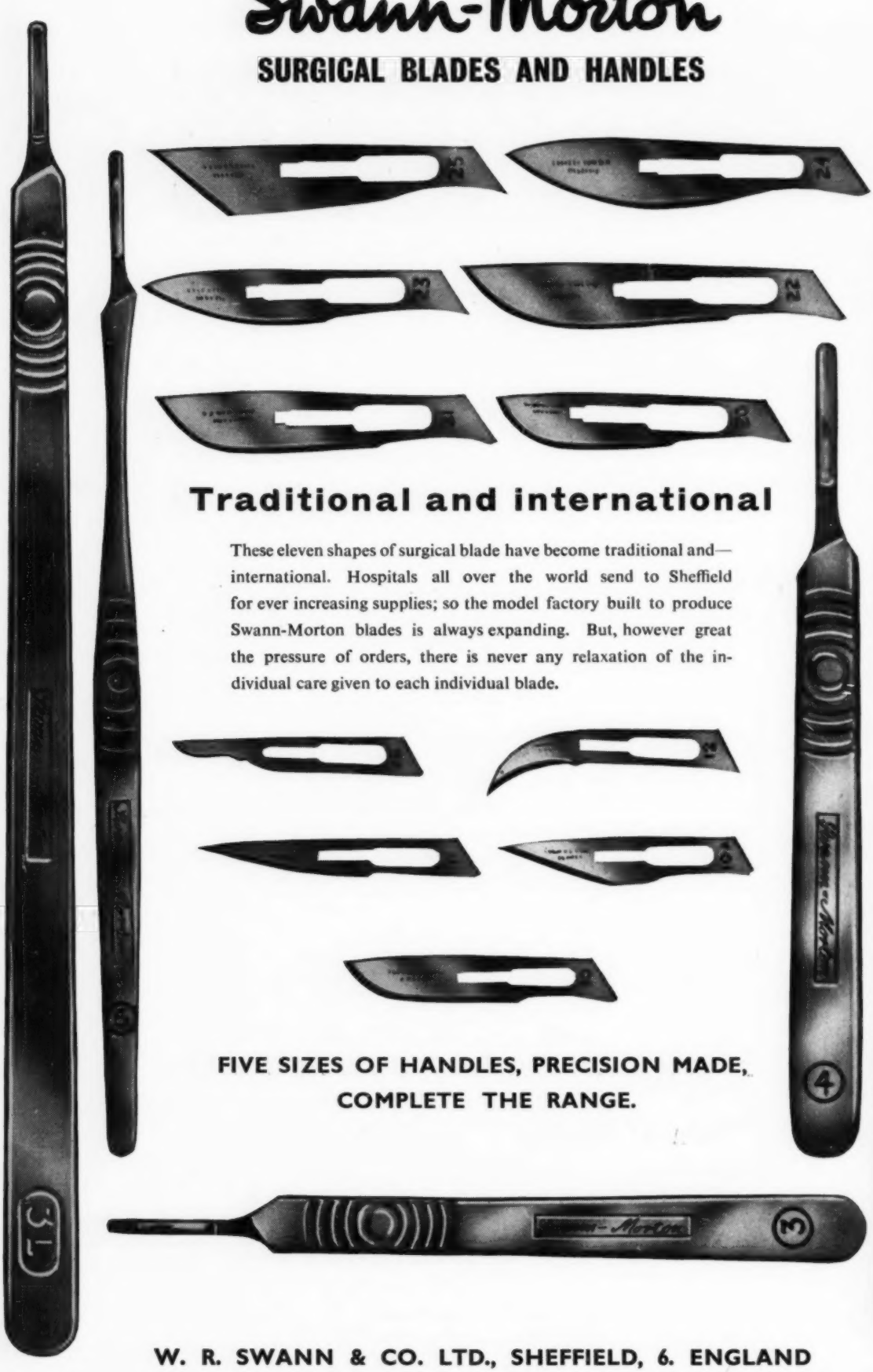

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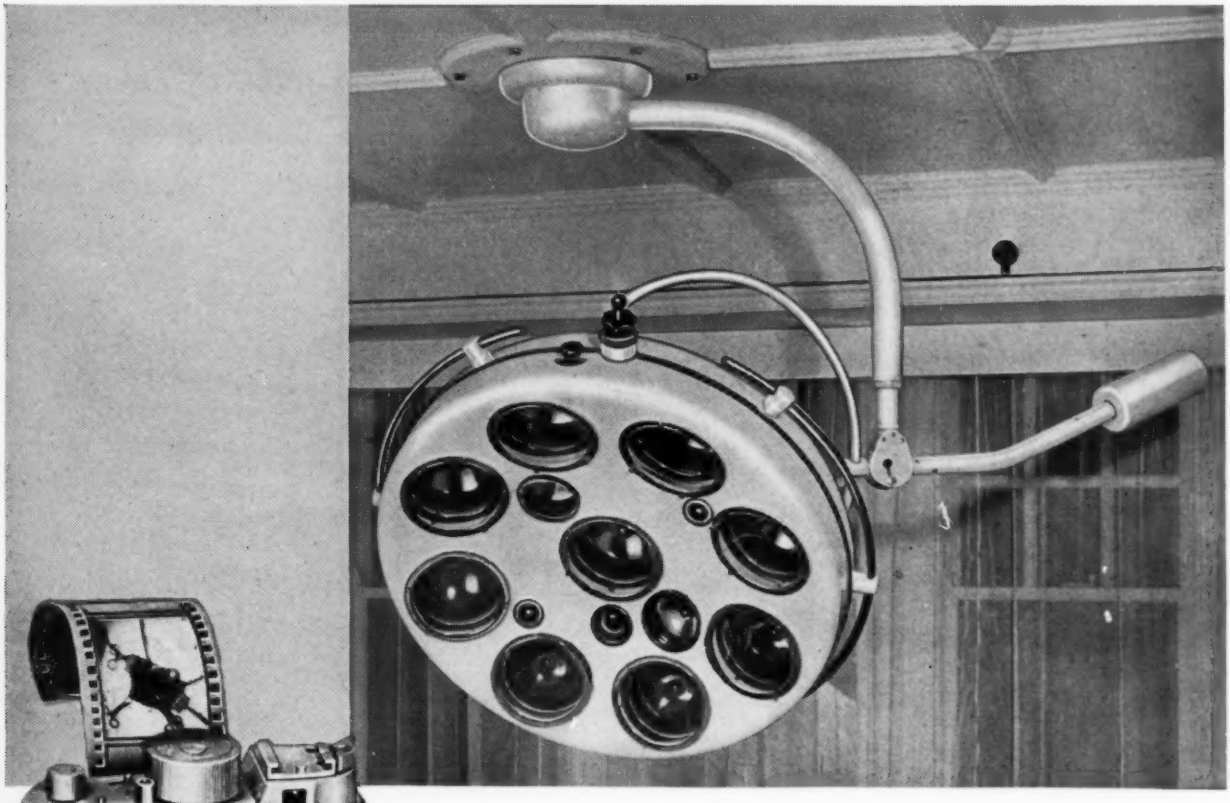

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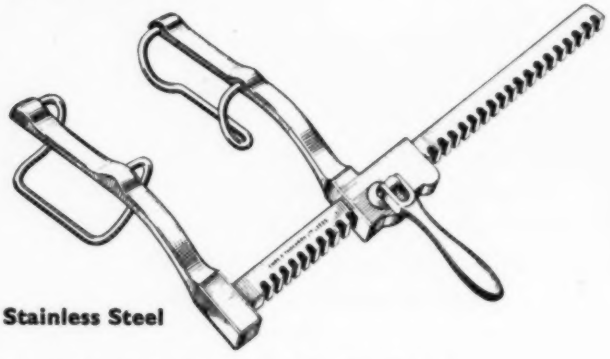

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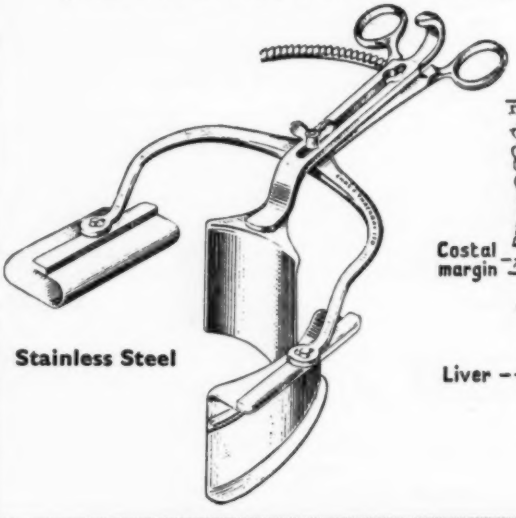

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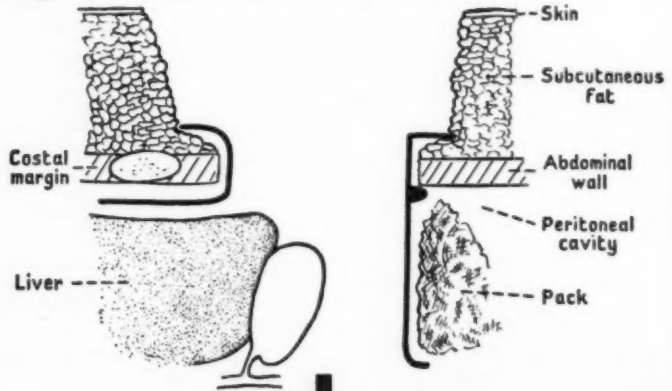

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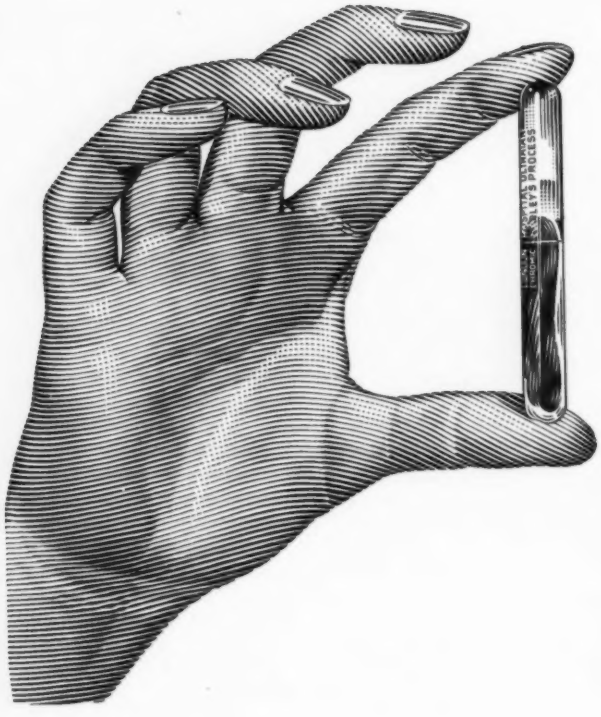

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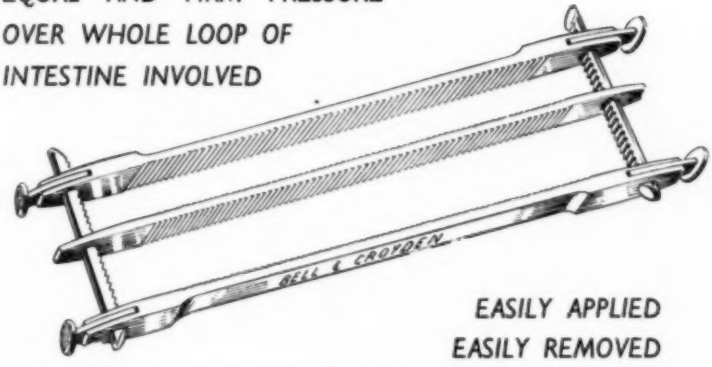

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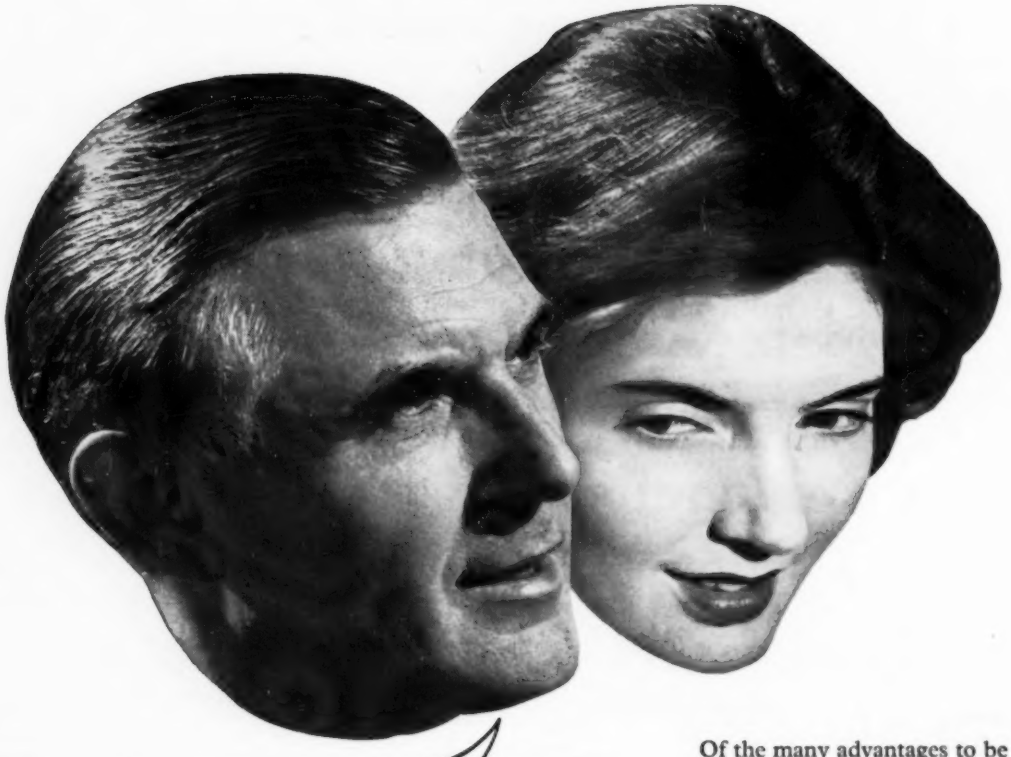

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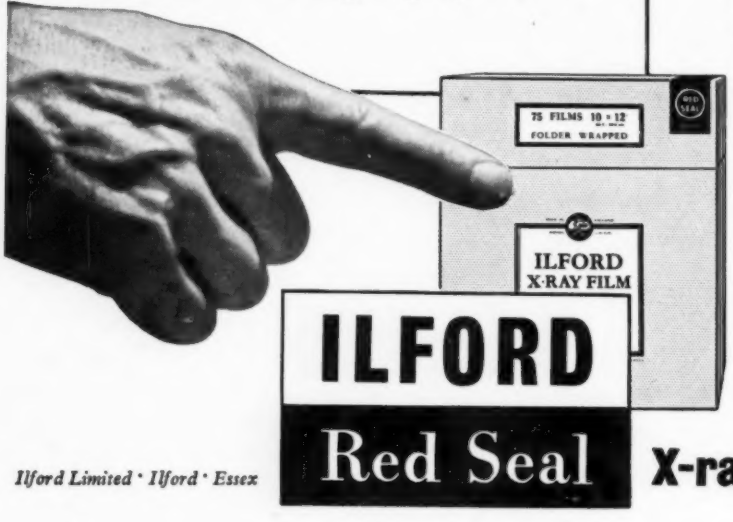

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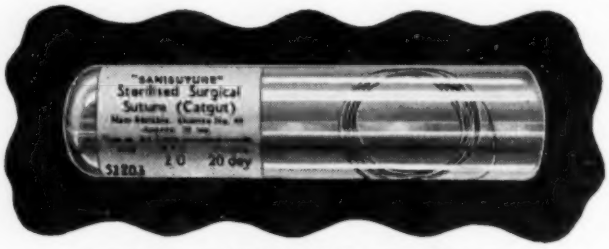

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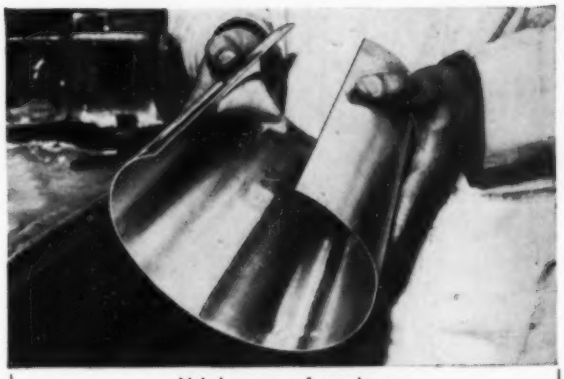
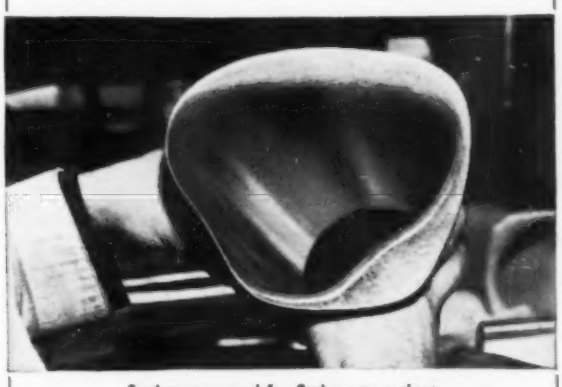
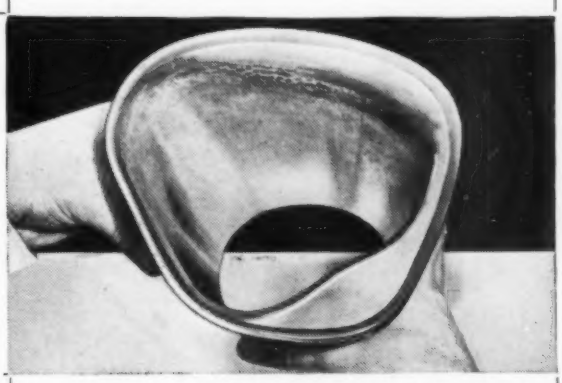


Alclad cone cut from sheet.

Preparing to shape anatomical features of socket.



Socket prepared for fittings to patient.



Socket after successful fittings, rimmed, and ready for assembly to leg.

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